Prevalence and impact of chronic diseases in adolescents with intellectual disability

Oeseburg, Barth

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Contents

Chapter 1

Introduction

1.1. Introduction

Information about the prevalence and impact of chronic diseases on adolescents with intellectual disability (ID-adolescents) is scarce. Schools for practical training and schools for special secondary education are in need of more knowledge about the chronic diseases of their pupils with intellectual disability (ID) and about the impact they have on their emotional and behavioural functioning.
Schools for practical training and schools for special secondary education prepare pupils to participate in society and to enter the labour market [1]. Therefore, information is needed on whether and to what degree ID-adolescents have additional chronic diseases. Both ID and chronic diseases can lead to limitations in adolescents' capacities. Capacities seem to be particularly limited in case of emotional and behavioural problems. In addition, limited capacities can have a profound effect on participation of ID-adolescents in educational programs and occupational opportunities [2-11].
However, evidence on the prevalence rates of chronic diseases in ID-adolescents and the impact on their emotional and behavioural functioning is limited.
In this chapter the key concepts of this thesis are introduced: intellectual disability, chronic diseases, and the impact of chronic diseases in ID-adolescents on their emotional and behavioural functioning. Moreover, the broader context of this thesis: the rights of the child, the EQUAL project Prevention and Prevalence, the Dutch education system and the occupational opportunities in the Netherlands for ID-adolescents will be described. At the end of this chapter, the aims of this thesis, the research questions, the methodology of the studies and outline of this thesis will be presented.

1.2. Intellectual disability

Intellectual disability is defined as a disability characterized by significant limitations both in intellectual functioning (reasoning, learning, problem solving) and in adaptive behaviour, which covers a range of everyday social and practical skills. This disability originates before the age of 18 [12].
Currently the term 'intellectual disability' is the preferred term above 'mental retardation' and 'learning disability'. This change in definition reflects the transition from a medical-individualistic approach to a social-ecological approach whereby a disability is defined in the broader context of the interaction between the person and his/her environment, and the personalized supports that are required by the individual to enhance this interaction [13, 14].

Based on the IQ distribution the estimation is that about 2% of people in the total population have IQs < 70 (two standard deviations below the norm), and 14% of people in the total population have IQs < 85 (one standard deviation below the norm) [15]. In the Netherlands there are about 3.9 million children aged 0-18 and estimates are that about 550.000 of them have an ID. This concerns 78.000 with an IQ < 70 and 472.000 with IQs between 70 and 84 [16].

1.3. Chronic diseases

A disease or health condition in children is considered to be chronic if the following criteria are met: (1) its diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods and instruments according to the professionals; (2) it is not (yet) curable or, for mental health conditions, it is highly resistant to treatment; (3) it has been present for longer than three months, or it will, very probably, last longer than three months or it has occurred three times or more during the past year and will probably recur again [17, 18].
Epidemiological research suggests that at least 12% of the adolescents in the general population live with a chronic disease, and this number may largely increase in the future [10, 18, 19]. There is growing evidence that adolescents with chronic diseases are doubly disadvantaged since adolescents with somatic or mental chronic diseases in the general population have, next to disease specific challenges, a higher risk on emotional and behavioural problems compared to their healthy peers [10, 20-31].

1.4. Chronic diseases in ID-adolescents and the impact on emotional and behavioural problems

Literature about young people with ID indicates that ID-adolescents have a greater risk on chronic diseases compared to their peers without ID, but conclusive evidence on the prevalence rates of a wide range of chronic diseases in ID-adolescents is lacking [6, 32-39]. In addition, ID-adolescents also have a higher risk on emotional and behavioural problems - including the full range of pervasive developmental disorder behaviour (PDD behaviour) - compared to their peers without ID [6, 40-46].
Whether the combination of ID and chronic diseases -number and nature- in adolescents leads to a higher risk for emotional and behavioural problems is

unknown. Evidence on the impact of chronic diseases in ID-adolescents on emotional and behavioural problems is highly needed. If occurring, the resulting accumulation of problems may provide additional reasons for early detection and treatment of behavioural problems among this group. Such early interventions may improve participation of ID-adolescents in educational programs and subsequently their occupational opportunities and societal participation [2-8, 11, 30, 40, 47-51]

1.5. Context of the thesis

1.5.1. The rights of the disabled child in international law

This thesis is situated in several international human rights instruments, in particular the United Nations Convention on the Rights of the Child (CRC) and the Convention on the Rights of Persons with Disabilities (CRPD). Both Conventions pay special attention to the legal position of children, i.e. adolescents with disabilities. The Dutch government has ratified CRC on February 6, 1995 and signed CRPD on March 30, 2007 [52]. The ratification of CRPD by the Dutch government is expected soon [53].

Several provisions of these human rights treaties are relevant for the legal position of the ID-adolescent. Article 23 section 1 CRC states that the mentally or physically disabled child is entitled to enjoyment of a full and decent life, in conditions which ensure dignity, promote self-reliance and facilitate the child's active participation in the community. Section 2 of this provision provides for the disabled child's right to special care and assistance from different resources extended to both the child and primary caregivers. Section 3 specifies the steps States Parties need to take in order to implement this right, in particular in the areas of education, training, health care rehabilitation services, preparation for employment and recreation opportunities, in a manner conducive to the child's achieving the fullest possible social integration and individual development. Finally, section 4 emphasizes the importance of international cooperation and the exchange of appropriate information in the field of preventive health care and medical, psychological and functional treatment of disabled children [54, 55].

With regard to the rights of ID-adolescents on education Article 28 CRC in relation to Article 2 section 1 CRC must be mentioned. Under these provisions children with disabilities have the same right to education as other children and are to achieve this right on the basis of equal opportunity and without discrimination of any kind, including disability [56]. Furthermore, Article 29 CRC defines fundamental

aims of the education of the child. Basically, a child's education must be targeted at the development of the child's personality, talents and mental and physical abilities to its fullest potential [54].

In relation to the transition from school to work, it must be stressed that career development is regarded as a continuous process throughout life. This process starts at elementary school and should be followed by a functional secondary school curriculum. Such a curriculum must stimulate the development of adequate skills and access to work (-experience), under systematic coordination and monitoring, at school as well as in the work place. In fact, the importance of employment for persons with disabilities is emphasized by CRPD (Article 27). This Article states that the right of persons with disabilities to work - on an equal basis with others - should be recognized by the States Parties. This includes the right to the opportunity to gain a living by work freely chosen or accepted in a labour market and to a work environment that is open, inclusive and accessible to persons with disabilities [57].

1.5.2. Prevention and Prevalence

This thesis is part of the Prevention and Prevalence project. The Prevention and Prevalence project started in 2005 and was funded by the European Social Fund (EQUAL Program part II). EQUAL was a laboratory for new ideas to the European employment strategy and the social inclusion process. Its mission was to promote a more inclusive work life through fighting discrimination and exclusion based on sex, racial or ethnic origin, religion or belief, disability, age or sexual orientation. The goals of the Prevention and Prevalence project were:

1. to explore the prevalence of chronic diseases in adolescents with intellectual disabilities (ID-adolescents) attending schools for practical training and special secondary schools and the impact of chronic diseases on their behaviour;
2. to assess the knowledge about chronic diseases in ID-adolescents among teachers working in schools for practical training and special secondary schools;
3. to support teachers to handle disease specific problems and emotional and behavioural problems in the educational program and the transition to work of these adolescents.

1.5.3. Education

This thesis is mainly focused on ID-adolescents attending schools for practical training and schools for special secondary education. In the Netherlands, every

child older than four years is legally obliged to attend school for at least 12 full school years and, in any event, until the end of the school year in which they turn 16. After this period of full-time compulsory schooling, children under 18 years must attend school at least one day a week until the end of the school year in which they turn 17 [1].
Next to mainstream primary schools there are special schools for children with disabilities and learning and behavioural difficulties who - at least temporarily - require special educational treatment. After leaving primary school (after eight years of primary school) children can choose between two types of secondary education:

1. pre-university education, senior general secondary education, pre-vocational secondary education which prepares pupils for university education, professional education and secondary vocational education, respectively;
2. schools for practical training for ID-adolescents (IQ between 60 and 80/85) who are unable to obtain a pre-vocational secondary education qualification which prepares them for entering the labour market.

As primary education, there are also special schools for secondary education in case adolescents with disabilities are not able to - temporarily - participate in mainstream schools. The aim of this kind of education is to enable as many pupils as possible to return to mainstream education. Adolescents attending special schools for secondary education who are not able to participate in mainstream schools, most of them with ID, are prepared for entering the labour market. Special schools for secondary education are divided into four categories:

1. schools for visually disabled adolescents with or without ID;
2. schools for adolescents with communication disabilities (due to hearing, language or speech difficulties) with or without ID;
3. schools for physically disabled adolescents or adolescents with ID (IQ < 60, or IQ between 59 and 70 with other severe disabilities);
4. schools for adolescents with psychiatric or behavioural disorders with or without ID.

Teachers working with ID-adolescents with or without chronic diseases are more challenged than their colleagues working with adolescents without disabilities. Both, ID and chronic diseases, affect educational success. The limited evidence shows that teachers' knowledge on chronic diseases in adolescents is far below

what is necessary. This can result in inaccurate appraisal of the adolescents' limitations, capacities and functioning at school [3, 4, 8, 49, 58]. Teachers need information on chronic diseases among ID-adolescents since it may help them to manage the effects of chronic diseases on the adolescents' functioning in educational programs, and to support them in the problematic transition from school to work [59].

1.5.4. Occupational opportunities

The combination of ID with chronic diseases and/or emotional and behavioural problems has a profound effect on the occupational opportunities of adolescents [2-11].

It is difficult to provide exact numbers of ID-adolescents entering the labour market because studies on this topic vary regarding methodologies, including the definition of the target group [60-62]. However, the estimation is that about 25-35% of all young people with a disability aged 15 to 22 years have a job on the labour market, against 77% of the young people in the total population [60]. In addition, people with ID have a lower chance to enter the labour market compared to their peers [61]. Moreover, a growing proportion of all youngsters receive disablement benefits under the Youth Employment Disability Benefits Act scheme (WAJONG), in particular those with ID. The WAJONG is established for adolescents leaving school and declared unfit or partially unfit for work. The WAJONG scheme ensures that these youngsters receive a minimum level income and provides several instruments for claimants (e.g. schooling, training) and employers (e.g. wage dispensation, rebate of contributions for disability and unemployment) that could be used to promote the participation in the labour process of these youngsters [63, 64].

The number of young adults aged 18-24 (n≈1.419.000 [16]) receiving a WAJONG benefit doubled between 2000 and 2008; from 24 thousand (1.7%) in 2000 to more than 49 thousand (3.5%) in 2008. Most claimants (85%) are pupils leaving practical training schools and secondary special schools with mental or psychological disability, rather than a physical disability [59, 60, 65]. On the other side, only 4% of the claimants leave the WAJONG yearly [66]. Moreover, the spending on the WAJONG increased dramatically from 1.3 billion euro in 2003 to 1.9 billion euro in 2008 [64, 67].

1.6. Aims of this thesis

The aims of this thesis are: (1) to explore the prevalence of chronic diseases in adolescents with intellectual disability and the impact on their behaviour; and (2) to assess the knowledge about chronic diseases in ID-adolescents among teachers working in schools for practical training and special secondary schools. These goals have been translated in the following five research questions:

1. What is known in the literature on the prevalence rates of chronic diseases in populations of children with ID?
2. What is the prevalence of chronic diseases in ID-adolescents aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people)?
3. What is the concordance between the knowledge teachers have on the presence of chronic diseases in ID-adolescents and the knowledge of parents and healthcare professionals?
4. What is the association between chronic diseases in ID-adolescents and their emotional and behavioural functioning?
5. What is the association between chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour?

1.7. Methods

1.7.1. Population and procedures

The data for this thesis were collected in 2006-2007 on adolescents with a borderline, mild, moderate, severe ID aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people).

These adolescents attended schools for practical training or special secondary schools. ID-adolescents attending schools for practical training can be classified as mainly educable and have IQs between 60 and 84. ID-adolescents attending special secondary schools can be classified as mainly trainable and have IQs between 30 and 59 [41]. All parents of the 2156 adolescents aged 12-18 years were asked to fill in a questionnaire on background characteristics, chronic diseases of their child and his/her emotional and behavioural functioning. In addition, in the prevalence study on chronic diseases also adolescents not attending school (n=115), most of them with IQs < 30, attending day care centres or living in

residential centres in the north of the Netherlands, Groningen and Drenthe, were included. The parents of these adolescents were asked to fill in a questionnaire on background characteristics and the presence of chronic diseases in their child. Finally, in the study regarding the concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents, teachers and the GPs of the adolescents attending schools for practical training or special secondary schools were asked to fill a questionnaire on background characteristics and the presence of chronic diseases in their pupil or patient, respectively.
Detailed information on the data collection: study selection, sample and subsamples and data analyses are described in the various methods sections of the chapters.

1.7.2. Measures

1.7.2.1. Chronic diseases

Data on the occurrence of chronic diseases in ID-adolescents were obtained by the National Permanent Survey on Living Conditions questionnaire (POLS), module health and labour, part chronic diseases in children [68]. POLS is periodically administered in a representative population sample (n≈10.000). To improve the validity POLS was revised in 1999 [69]. Questions were adjusted and added for example about the presence of prevalent chronic diseases.
POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic systems and Attention Deficit Hyperactivity Disorder (ADHD). To cover also prevalent chronic diseases in adolescents with ID questions were added about the presence of pervasive developmental disorders (PDD). Parents were asked to fill in the presence or absence of each specific chronic disease in the last twelve months for their child. Parents were also offered the possibility to mention chronic diseases that were not listed in the questionnaire. In the study regarding the concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents, teachers and GPs of the adolescents were asked to fill in the same questionnaire as the parents.

1.7.2.2. Emotional and behavioural functioning

Emotional and behavioural problems - including the full range of pervasive developmental disorder behaviour (PDD behaviour) - in ID-adolescents were assessed by the validated Dutch parent version of the Strengths and Difficulties

Questionnaire (SDQ) [70-72] and the Dutch parent version of the Children's Social Behaviour Questionnaire (CSBQ) [40, 43, 73, 74].

The SDQ is a world wide used screening instrument for emotional and behavioural problems: conduct problem; inattention-hyperactivity; peer problem and prosocial behaviour. Several studies have shown the good reliability and validity of the SDQ in a non-ID population [75, 76].

The SDQ has been developed for youth without ID, but it is also used in studies with children and adolescents with ID to measure their emotional and behavioural functioning [44, 77-80]. Kaptein et al. [44] found that the internal consistency of the SDQ scales was comparable to the internal consistency in a sample without ID. Recently the SDQ is implemented in the Dutch Preventive Child Health Care. SDQ in particular the total difficulties scale added most to the identification of psychosocial problems by the preventive child healthcare professionals compared to other questionnaires. Moreover, SDQ is short, easy to answer and - due to the limited time available for a standard health examination - easy to score [70, 81].

The CSBQ has been developed in the Netherlands as a screening instrument for children with pervasive developmental disorder (PDD). The CSBQ describes a broad range of PDD behaviour, including milder forms that are typical of pervasive developmental disorder: not optimally tuned to the social situation; reduced contact and social interest; orientation problems in time, place, or activity; difficulties in understanding of social information; stereotyped behaviour; and fear of and resistance to changes. The CSBQ was originally developed for and investigated in children with normal intelligence to measure (sub-threshold) autistic symptoms. In that group the psychometric qualities of the CSBQ were reported to be good. Subsequent research shows that this also holds for children with ID [73, 82].

1.8. Outline of the thesis

Chapter 2 provides a systematic literature review on the prevalence rates of chronic diseases in populations of children with ID. Chapter 3 shows the prevalence rates of chronic diseases in ID-adolescents aged 12 – 18 in two provinces in the north of the Netherlands, Groningen and Drenthe (total population about 1.1 million people). In addition, these prevalence rates were compared with the prevalence rates among adolescents in the general population. In Chapter 4 the knowledge of teachers on the presence of chronic diseases in ID-adolescents is compared with the knowledge of parents and healthcare professionals. Chapter 5 focuses on

the impact of chronic diseases in ID-adolescents on the increase of the likelihood of emotional and behavioural problems. Chapter 6 focuses on the impact of chronic diseases in ID-adolescents on PDD behaviour, in particular the association between somatic chronic diseases and milder forms of PDD behaviour. Finally, in Chapter 7 the main results are summarized and discussed. Practical and research implications are given at the end.

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Chapter 2

The prevalence of chronic diseases in children with intellectual disability: a systematic literature review

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Abstract

The aim of this study is to provide a systematic review of the prevalence rates of chronic diseases in populations of children with intellectual disability (ID) in general. We identified 2994 relevant studies by searching Medline, Cinahl and PsycINFO databases in the period 1996-2008. Finally 31 studies had a sufficient methodological quality and were included. The six most prevalent chronic diseases in children with ID were epilepsy (22.0%), cerebral palsy (19.8%), any anxiety disorder (17.1%) oppositional defiant disorder (12.4%), Down Syndrome (11.0%) and autistic disorder (10.1%). The reported prevalence rates of chronic diseases in children with ID are much higher than in the general population. However, both the number of studies that were included and the number of chronic diseases they reported about were limited. There is an urgent need for better evidence on the prevalence of chronic diseases in children with ID.

2.1. Introduction

The past decades have shown an increase in the knowledge on chronic diseases in children with intellectual disability (ID). In several studies ID in children was associated with a range of chronic diseases [1-5].

A major difficulty in studies on this subject is the wide variability of prevalence rates that are reported for specific chronic diseases in children with ID. This variability is a result of heterogeneity regarding several factors, such as size, definition and recruitment of the study population, response rate, method of diagnosis and the use of different classification frameworks for diagnosing certain disorders in children with ID. As a result, the validity of prevalence rates has to be disputed [6-9].

Both policymakers and professionals value and need valid prevalence rates. Policymakers need these data for the planning and financing of adequate care arrangements (e.g. health, education, work) in order to enhance the well-being and societal participation of children with ID and their families. Professionals need these data for the early detection and adequate treatment of chronic diseases and to prevent the burden of these conditions in ID-adolescents and their families [2, 10-16].

The aim of this study is to provide an overview of the prevalence rates of chronic diseases in populations of children with ID in general by performing a systematic literature review. This review is part of a research project on chronic diseases in a population of children with ID in general in the Netherlands.

2.2. Methods

2.2.1. Data sources / Study identification

The following electronic databases were searched for relevant studies in the period 1996-2008: Medline, Cinahl and PsycINFO. Search terms were related to the exploded terms: mental retardation, intellectual disability, mentally disabled persons, mental development, intellectual development, developmental disability or disabilities, learning disability or disabilities, or learning development in Major Medical Subject Headings Descriptor (MJME) and learning disorders in all subheadings. We used the ICD-9 classification chapter entitled 1 to 16 [17], the ICD-10 classification chapter entitled I to XVIII [18], the DSM III and III-R classification chapter entitled “Disorders Usually First Evident In Infancy, Childhood, Or Adolescence” [19, 20], and the DSM IV and IV-TR classification chapters entitled “Disorders Usually First Diagnosed in Infancy, Childhood, or Adolescence” [21, 22] for the

selection of chronic diseases in children. The diagnostic titles were translated in Medical Subject Headings (MeSh) with complications, diagnosis or epidemiology in all subheadings. Additional studies were obtained from the reference lists of the included studies.

2.2.2. Study selection

In order to be considered for inclusion, studies had to report mainly about children (<19 years) diagnosed with borderline, mild, moderate, severe or profound ID and with a chronic disease. ID was ideally established by a validated IQ - and / or adaptive behaviour test and ideally classified according to the Diagnostic and Statistical Manual of Mental Disorders classifications (DSM) [19-22] or the International Statistical Classification of Diseases (ICD) [17, 18]. Although borderline ID has not been an accepted level of intellectual disability for quite a long time, studies reporting on children with borderline ID were included because these group faces substantially elevated cognitive and morbidity risks and also faces problems in adaptive behaviour [23, 24].

A chronic disease was defined as a chronic physical, developmental, behavioural, or emotional condition in children aged 0 up to 18 years. The diagnosis had to have been present for longer than three months or if it would, very probably, last longer than three months, or had to have occurred three times or more during the past year and would probably recur. The chronic disease had to be diagnosed according to professional standards or extracted from medical case files or registers and ideally classified according to the DSM or ICD [15, 17-22, 25].

Eligible studies were English language studies that had a cohort, patient-control or cross-sectional design and a focus on a population of children with ID in general. We excluded studies that focused exclusively on subpopulations of children with a specific biomedical cause of ID such as Down syndrome because reviews are already available about the prevalence of chronic diseases in these children [26-28]. In addition, studies on only adults, studies with other designs (e.g. case studies), studies not reporting about original research (e.g. reviews or comments) and studies in which the diagnoses were based on screening instruments were excluded.

Three reviewers, BO, GD and DJ independently screened the titles and abstracts of the relevant studies in the period 1996-2008 for eligibility. Each reviewer screened 2/3 of the titles and abstracts; so each title and abstract was screened by two reviewers. Any discrepancies between reviewers were resolved by discussion among the reviewers, if necessary by obtaining the full text article and by consultation of a fourth reviewer, SR.

The inter-rater reliability statistics were very good [29]. Cohen's weighted kappas were 0.81 (GD-DJ), 0.82 (BO-DJ) and 0.82 (BO-GD) for the screening of titles and abstracts (outcomes: inclusion, exclusion or doubt). There was disagreement among the reviewers in 8 to 10% of the 2994 studies. Most disagreements were resolved by discussion among the reviewers. In twelve unsure cases the full text of the article was obtained because eligibility could not be determined from the title and abstract alone. In two cases of remaining doubt, the fourth reviewer (SR) was consulted.

2.2.3. Data extraction

Full papers of potentially eligible studies were obtained and independently analyzed by the three reviewers. Teams of two reviewers independently assessed all studies identified for full-text analysis; BO analyzed all the studies, whereas GD and DJ analyzed about half of the eligible studies. The reviewers used a structured data form based on the Strengthening The Reporting of OBservational studies in Epidemiology (STROBE) checklist to extract the key data for the assessment of the methodological quality of the included studies [30].

In 570 studies eligible for methodological quality assessment (values: low, weak to medium, good and high) disagreement among the reviewers was found in 16 to 17% of the studies. However, Cohen's weighted kappas were very good, 0.85 (BO-DJ) and 0.89 (BO-GD) [29], because the inter-rater disagreements were smaller compared to the inter-rater disagreements of the screening of titles and abstracts. All disagreements were resolved by discussion between the reviewers.

2.2.4. Assessment of methodological quality

There is not a 'gold standard' for assessing the quality of observational studies [31]. The reviewers consequently developed a method based on the domains that are of importance for the internal and external validity of the studies. The methodological quality of the eligible studies was assessed by examining the following methodological characteristics: contextual information, population (bias), selection (bias), outcome measures reliability, results and confounders. The reviewers assigned weights to each characteristic taking into account the importance of each characteristic for the internal and external validity [31-33]. The appraisal of the overall methodological quality of the individual studies was based on a total score on the different indicators of these characteristics (Table 1).

The total quality scores for each study were calculated by summing up the score on each indicator and could range from 0 to 11 points. We classified the quality scale scores into the following ordinal categories: low quality (score < 6), weak to

medium quality (score 6-7), good quality (score 8 -9) and high quality (score 10-11) [34, 35]. Studies with a low methodological quality were excluded from further analyses.

Table 1: Indicators and score range of the methodological characteristics.

Traits	Indicators	Score
Contextual information	Description of the study (design, ID levels, chronic disease & socio demographic characteristics)	0-1
Population (bias)	In-and exclusion criteria	0-3
Response (bias)	Response rate	0-3
Outcome measures reliability	Diagnosis of ID and chronic disease according to professional standards or extracted from medical case files or registers	0-2
Results	Results and control for confounders	0-2

2.2.5. Analysis and synthesis

We first assessed inter-rater reliability by calculating Cohen's weighted kappas with MEDCALC v9.2.0.2. for the screening of titles and abstracts (BO-GD, BO-DJ, GD-DJ) and for the methodological quality assessment (BO-DJ, BO-GD) [29]. Subsequently, the prevalence rates of the included studies were grouped by: somatic chronic diseases (epilepsy, cerebral palsy, visual problems, hearing problems and miscellaneous somatic chronic diseases); congenital malformations (genetic-chromosomal / sex-linked malformations and other chromosomal, endocrine / metabolic diseases malformations); pervasive developmental disorder (autistic disorder (AD), pervasive developmental disorder (PDD) other than AD); attention-deficit/hyperactivity disorder (ADHD) / hyperkinetic disorder; and miscellaneous psychiatric disorders. The weighted mean prevalence rate and the 95% confidence intervals were calculated if more than two studies reported about a disease. The weighted mean is used to aggregate the prevalence rates of the different chronic diseases that were found in each study to a single resultant score. Studies with large populations contribute more to the weighted mean than smaller ones (Figure 1).

$$\text{Weighted mean} = \frac{\Sigma\ (\text{prevalence rate X number of respondents})}{\Sigma\ (\text{number of respondents})}$$

Figure 1: Formula weighted mean

2.2.6. Search for trials

Figure 2 shows the study selection process. The database search resulted in 3216 potentially relevant studies: 2478 Medline, 510 Cinahl, and 228 PsycINFO; 241 of these were duplicates. The screening of the reference lists of the included studies added another 19 studies leading to a total number of 2994 studies. After screening the titles, 2424 studies were excluded because they did not meet the inclusion criteria. After full-text analysis 539 studies were excluded because of the following main reasons: study reported exclusively about subpopulations of children with a specific biomedical cause of ID (n=170), selective population - or low response rate (n=107), study did not report about chronic disease or ID (n=92), review, case-report or comment / letter (n=82), age-range too broad (n=65), diagnoses based on screening instruments (n=23). Finally, 31 studies were included (Table2).

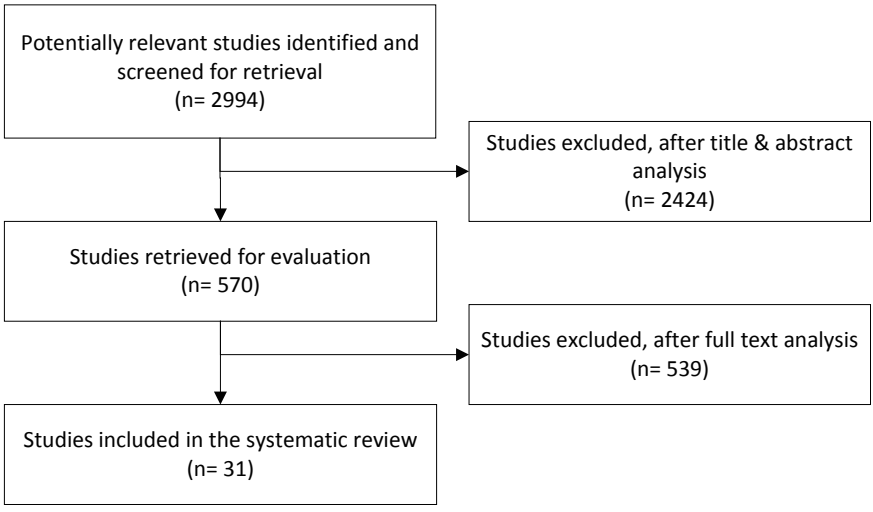

Figure 2: Flow diagram of study selection process

Table 2: Characteristics of the included studies.

Study	Quality	Country	Chronic disease(s)	Level of ID	Age group	Number of subjects with ID (response %)	Recruitment Population with ID
Nordin et al. (1996)[36]	good	Sweden	epilepsy, CP, AD, PDD other than AD	mild-profound	3-18	101 (100%)	special schools & rehabilitation centres
Steffenburg et al. (1996)[37]	good	Sweden	epilepsy	mild-profound	8-16	378 (100%)	paediatric (outpatient) clinics, psychiatric (outpatient) clinics and rehabilitation centre
van Schrojenstein Lantman et al. (1997)[38]	good	Netherlands	epilepsy, CP, visual problems, hearing problems, miscellaneous somatic chronic diseases, miscellaneous psychiatric disorders	mild-profound	0-19	1026 (72%)	residential centres
Yeargin-Allsop et al. (1997)[16]	high	USA	miscellaneous somatic chronic, congenital malformations	mild-profound	10-12	715 (95%)	schools, health - and social services
Hou et al. (1998)[11]	high	Taiwan	miscellaneous somatic chronic, congenital malformations	mild-profound	6-18	11892 (100%)	schools, outpatient clinic and institutions
Fernell (1998)[39]	good	Sweden	epilepsy, CP, hearing problems, congenital malformations, AD, PDD other than AD	moderate-profound	3-16	64 (100%)	paediatric (outpatient) clinic and social services
Cans et al. (1999)[40]	good	France	CP, miscellaneous somatic chronic diseases, congenital malformations	moderate-profound	7-16	1150 (100%)	special schools
Airaksinen et al. (2000)[41]	high	Finland	epilepsy, CP, congenital malformations, PDD other than AD	mild-profound	0-18	151 (85%)	schools and social services
Lewis et al. (2000)[42]	good	Australia	epilepsy	mild-profound	8-22	392 (100%)	schools, health - and social services
Merrick et al. (2000)[43]	good	Israel	HIV	mild-profound	0-18	1321 (100%)	schools, health - and social services
Stromme et al. (2000a)[14]	high	Norway	epilepsy, CP, miscellaneous somatic chronic diseases, congenital malformations	mild-profound	8-13	178 (96%)	special schools and health services
Stromme et al. (2000b)[44]	high	Norway	AD, PDD other than AD, hyperkinetic disorder, miscellaneous psychiatric disorders	mild-profound	8-13	178 (96%)	special schools and health services
Magnusson et al. (2001)[45]	weak to medium	Iceland	AD, PDD other than AD	mild-profound	5-14	not given	hospital and diagnostic centre for PDD
Molteno et al. (2001)[46]	weak to medium	South Africa	epilepsy, CP, AD, ADHD	mild-profound	6-18	355 (100%)	special schools & training centre

Christianson et al. (2002)[47]	high	South Africa	epilepsy, CP, hearing problems, congenital malformations	borderline-profound	2-9	238 (100%)	private households
Koskentausta et al. (2002)[48]	high	Finland	epilepsy, AD, PDD other than AD, hyperkinetic disorder, miscellaneous psychiatric disorders	mild-profound	6-13	155 (100%)	special schools, hospitals and rehabilitation centre
Dekker et al. (2003)[49]	good	Netherlands	epilepsy, congenital malformations, ADHD, miscellaneous psychiatric disorders	borderline-moderate	6-18	474 (87%)	special schools
Emerson (2003)[50]	high	UK	hyperkinetic disorder, miscellaneous psychiatric disorders	not given	5-15	264 (83%)[a]	private households
Jeliffe-Pawlowski et al. (2003)[51]	high	USA	epilepsy, CP	mild-profound	7-9	613 (100%)	department of developmental services
Morgan et al. (2003)[52]	high	UK	epilepsy	mild-profound	0-19	258 (100%)[b]	hospitals and outpatient clinics and social services
de Bildt et al. (2005)[53]	high	Netherlands	AD, PDD other than AD	mild-profound	4-18	825 (78%)	special schools, day-care - and residential centres
Zhang (2005)[54]	high	China	CP, visual problems, hearing problems, miscellaneous somatic chronic diseases, congenital malformation, AD	mild-profound	2-6	79 (100%)	community registers
Bradley et al. (2006a)[55]	good	Canada	AD	mild-profound	14-20	171 (67%)	schools, health - and social services
Bradley et al. (2006b)[56]	good	Canada	AD	mild-profound	14-20	171 (67%)	see Bradley et al. (2006a)
Voigt et al. (2006)[57]	good	USA	ADHD	borderline-mild	6-18	70 (100%)	schools and primary and special medical care providers
Emerson et al. (2007)[58]	high	UK	hyperkinetic disorder, miscellaneous psychiatric disorders	not given	5-16	641 (76%)[a]	see Emerson (2003)
Nielsen et al. (2007a)[59]	high	Denmark	visual problems	mild-profound	4-15	923 (82%)	schools en paediatric clinics
Nielsen et al. (2007b)[60]	high	Denmark	epilepsy, CP, visual problems, hearing problems	mild-profound	4-15	923 (82%)	see Nielsen et al. (2007a)
Petterson et al. (2007)[61]	high	Australia	congenital malformations	mild-profound	1-18	6106 (100%)	schools, health - and social services
Bryson et al. (2008)[62]	good	Canada	AD	mild-profound	14-20	171 (67%)	see Bradley et al. (2006a)
Gothelf et al. (2008)[63]	good	Israel	ADHD, miscellaneous psychiatric disorders	mild-moderate	12-21	87 (87%)	special schools

[a] Response rate in the sub sample of children and adolescents with ID is not available, instead we used the response rate of the total population children and adolescents in the survey

[b] We used a subset of the data that was provided to us by the authors

Abbreviations diseases: AD=Autistic Disorder, ADHD= Attention Deficit/Hyperactivity Disorder, CP=Cerebral Palsy, HIV=Human Immunodefiency Virus, PDD=Pervasive Developmental Disorder, PDD other than AD=Pervasive Developmental Disorder (PDD) other than autistic disorder (AD)

2.3. Results

2.3.1. Description of the studies

In Table 2, the characteristics of the 31 included studies that were left for analysis and synthesis are presented. Sixteen studies were classified as methodologically high quality studies, 13 as good quality studies and two as weak to medium quality studies. The studies were predominantly conducted in Europe (n=17). Six studies were conducted in North America, two in (South) Africa, two in Australia, two in Asia, and two in Israel. Most of the studies (n=22) focused on more than one chronic disease. The studies focused on the following chronic diseases: epilepsy (n=14), cerebral palsy (n=11), visual problems (n=4), hearing problems (n=5), miscellaneous somatic chronic diseases (n=7), congenital malformations genetic-chromosomal / sex-linked (n=10), congenital malformations other chromosomal, endocrine / metabolic diseases (n=9), autistic disorder (AD) (n=11), pervasive developmental disorder (PDD) other than AD (n=7), attention-deficit/hyperactivity disorder (ADHD) / hyperkinetic disorder (n=8), and miscellaneous psychiatric disorders (n=7). All studies reported about children (< 19 years). However, seven studies also included young adults with a maximum age of 22.

There was a huge variation in the size of the population with ID surveyed (range 64–11,892). Response rates were generally high (67% - 100%); only six studies had a response rate below 80%.

In most studies the children were recruited from (special) schools and certain health or social services: (special) schools and health services (n=8), (special) schools, health - and social services (n=7), (special) schools (n=3), private households (n=3), health - and social services (n=2), health services (n=2), (special) schools and social services (n=1), (special) schools day-care and residential centres (n=1), (special) schools and training centre (n=1), residential centres(n=1), community registers (n=1) and department of developmental services (n=1).

2.3.2. Methods of diagnosis and the classification framework

In Table 3 the various methods used in the assessment of the diagnoses of ID and of chronic diseases are presented as well as their classification frameworks.

2.3.2.1. Intellectual disability

In 20 studies ID was assessed by IQ tests, in 14 studies in combination with developmental tests. Six studies included children and adolescents that were eligible for health, education and social services for people with ID. In two studies the diagnosis ID was based on primary carer reports and/or teacher reports. One study included children and adolescents eligible for social services for people with ID or diagnosed with ID by school achievement and/ psychological tests, one study used developmental tests for establishing ID and in one study the ID diagnosis was extracted from the family history and medical records.

2.3.2.2. Chronic diseases

In 17 studies the diagnoses of chronic diseases was made by clinical examination or extracted from registers, medical files or from blood samples. Fourteen studies used multiple methods of diagnosis, in different combinations, for establishing the diagnosis. Most studies focusing on somatic chronic diseases or congenital malformations did not use a classification system for the diagnoses they reported. The International League Against Epilepsy (ILAE) criteria were used in three of the 14 studies reporting about epilepsy. The International Association for Prevention of Blindness (IAPB) criteria were used in one of the four studies reporting about visual problems. Just three studies used the ICD 9 or 10 as a classification system for different somatic chronic diseases or congenital malformations. The DSM or the ICD were used in most studies about PDD or psychiatric disorders. Four studies did not use a classification system for the PDD or psychiatric disorders diagnoses they reported.

2.3.3. Chronic diseases

Table 3 presents the prevalence rates of chronic diseases in the included studies and the methods of diagnosis and classification framework that were used. The weighted mean prevalence rate and the 95% confidence interval (CI) of the chronic diseases that were reported by more than two studies are presented in Table 4.

2.3.3.1. Epilepsy

Prevalence rates of epilepsy were reported in 14 studies and ranged from 5.5%-35.0%. The weighted mean prevalence rate was 22.0/100 (CI 20.8-23.2).

Table 3: Prevalence rates (in italics) of chronic diseases, methods of diagnosis and classification framework[a].

Epilepsy Study	Prevalence % (number/ respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Nordin et al. (1996)	22.0% (22/101)	IQ tests	not given	medical files	not given
Steffenburg et al. (1996)	26.0% (98/378)	IQ and DQ tests	not given	register	ILAE
van Schrojenstein Lantman et al. (1997)	27.0% (27/101)	eligibility for residential centres for ID	not given	medical files	ICD 9
Fernell (1998)	26.6% (17/64)	IQ and DQ tests	not given	medical files	not given
Airaksinen et al. (2000)	21.0% (32/151)[b]	school achievement / psychological tests or eligibility for social services for ID	ICD 10	clinical examination	ILAE
Lewis et al. (2000)	29.0% (115/392)	IQ tests	DSM IV	clinical examination	not given
Stromme et al. (2000a)	19.7% (35/178)	IQ and DQ tests	DSM IV	clinical examination	ILAE/ICD 10
Molteno et al. (2001)	23.7% (84/355)	eligibility for special schools for ID	not given	not given	not given
Christianson et al. (2002)	15.5% (37/238)	IQ and DQ tests	not given	clinical examination	not given
Koskentausta et al. (2002)	35.0% (55/155)	IQ and DQ tests	ICD 10	medical files	not given
Dekker et al. (2003)	5.5% (26/474)	eligibility for special schools for ID	not given	not given	not given
Jeliffe-Pawlowski et al. (2003)	26.1% (160/613)	IQ and DQ tests	not given	medical files	not given
Morgan et al. (2003)	27.9% (72/258)[c]	eligibility for social services for ID	ICD 9/ICD 10	register	ICD 9 or 10
Nielsen et al. (2007b)	19.5% (163/838)	IQ and DQ tests	not given	register	not given

Cerebral Palsy Study	Prevalence % (number/ respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Nordin et al. (1996)	8.9% (9/101)	IQ test	not given	medical files	not given
van Schrojenstein Lantman et al. (1997)	29.0% (29/101)	eligibility for residential centres for ID	not given	medical files	ICD 9
Fernell (1998)	23.4% (15/64)	IQ and DQ tests	not given	medical files	not given
Cans et al. (1999)	23.4% (269/1150)	IQ test	ICD 10	medical files	not given
Airaksinen et al. (2000)	10.6% (16/151)	school achievement / psychological tests or eligibility for social services for ID	ICD 10	clinical examination	not given
Stromme et al. (2000a)	14.0% (25/178)	IQ and DQ tests	DSM IV	clinical examination	ICD 10
Molteno et al. (2001)	33.8% (120/355)	eligibility for special schools for ID	not given	not given	not given
Christianson et al. (2002)	8.4% (20/238)	IQ and DQ tests	not given	clinical examination	not given
Jeliffe-Pawlowski et al. (2003)	18.4% (113/613)	IQ and DQ tests	not given	medical files	not given
Zhang (2005)	20.3% (16/79)	DQ tests	ICD 10	clinical examination	not given
Nielsen et al. (2007b)	15.9% (133/838)	IQ and DQ tests	not given	register	not given

[a] A list with an explanation of the different abbreviations is provided at the end of the table.
[b] The result is based on a cohort study when the respondents were 22 years. In a figure about the cumulative probability of developing epilepsy in the article we had the possibility to make an eyeball estimation when the respondents were 18 years. There was no difference between the values at 18 and 22 years.
[c] We used a subset of the data that was provided to us by the authors.

Table 3: (continued).

Visual problems					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
van Schrojenstein Lantman et al. (1997)	15.2% (15/101) visual impairment	eligibility for residential centres for ID	not given	medical files	ICD 9
Zhang (2005)	5.1% (4/78) visual impairment	DQ tests	ICD 10	clinical examination	not given
Nielsen et al. (2007a)	10.5% (97/923) visual acuity impairment 2.2% (20/923) visual field affected	IQ and DQ tests	not given	clinical examination	IAPB
Nielsen et al. (2007b)	15.3% (136/886) hyperopia 10.8% (96/886) myopia 20.6% (182/886) astigmatism 7.2% (64/886) anisometropia 26.8% (224/915) strabismus	IQ and DQ tests	not given	clinical examination	IAPB

Hearing problems					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
van Schrojenstein Lantman et al. (1997)	6.9% (7/101) hearing impairment	eligibility for residential centers for ID	not given	medical files	ICD 9
Fernell (1998)	4.7% (3/64) hearing impairment	IQ and DQ tests	not given	medical files	not given
Christianson et al. (2002)	7.1% (17/238) auditory disability	IQ and DQ tests	not given	clinical examination	not given
Zhang (2005)	0.0% (0/79) hearing disability	DQ tests	not given	clinical examination	not given
Nielsen et al. (2007b)	3.8% (32/838) hearing impairment	IQ and DQ tests	not given	register	not given

Miscellaneous somatic chronic diseases					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
van Schrojenstein Lantman et al. (1997)	8.9 % (9101) COPD 6.9% (7/101) gastric & oesophageal diseases 6.9% (7/101) back & neck disorders 4.0% (4/101) other chronic diseases	eligibility for residential centres for ID	not given	medical files	ICD 9
Yeargin-Allsop et al. (1997)	0.3% (2/715) cerebrovascular accident	IQ test	DSM III	medical files	not given
Hou et al. (1998)	2.2% (263/11892) cerebral/ intracranial hemorrhage 0.3% (41/11892) Reye syndrome	family history/ medical records/ blood and urine tests	not given	clinical evaluation/ blood sample	not given
Cans et al. (1999)	1.5% (17/1150) cerebral haemorrhage	IQ test	ICD 10	medical files	not given
Merrick et al. (2000)	0.0% (0/1321) human immunodeficiency virus	eligibility for social and educational services for ID	not given	blood sample	not given
Stromme et al. (2000a)	0.6% (1/178) cerebrovascular accident	IQ and DQ tests	DSM IV	medical files	not given
Zhang (2005)	2.5% (2/79) cerebrovascular accident 2.5% (2/79) osteoarthropathia	DQ tests	ICD 10	clinical examination	not given

Table 3: (continued).

Congenital malformations genetic-chromosomal and sex-linked					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Yeargin-Allsop et al. (1997)	4.8% (34/715) Down syndrome 0.1% (1/715) fragile-x 0.1% (1/715) Cri-du-Chat 0.3% (2/715) other than Down syndrome	IQ test	DSM III	medical files	not given
Hou et al. (1998)	13.1% (1557/11892) Down syndrome 2.0% (233/11892) fragile-x 0.2% (26/11892) Cri-du-Chat 3.3% (398/11892) other than Down syndrome	family history/ medical records/ blood and urine tests	not given	clinical evaluation, blood sample	not given
Fernell (1998)	20.3% (13/64) Down syndrome	IQ and DQ tests	not given	medical files	not given
Cans et al. (1999)	16.1% (185/1150) Down syndrome 2.3% (26/1150) other than Down syndrome	IQ test	ICD 10	medical files	not given
Airaksinen et al. (2000)	17.2% (26/151) all chromosomal	school achievement / psychological tests or eligibility for social services for ID	ICD 10	clinical examination	not given
Stromme et al. (2000a)	9.6% (17/178) Down syndrome 1.7% (3/178) fragile-x 1.1% (2/178) Cri-du-Chat 3.9% (7/178) other than Down syndrome	IQ and DQ tests	DSM IV	medical files	not given
Christianson et al. (2002)	2.1% (5/238) Down syndrome	IQ and DQ tests	not given	clinical examination	not given
Dekker et al. (2003)	5.3% (25/ 474) Down syndrome	eligibility for special schools for ID	not given	not given	not given
Zhang (2005)	2.5% (2/79) all chromosomal	DQ tests	ICD 10	clinical examination	not given
Petterson et al. (2007)	7.3% (446/6106) Down syndrome 3.0% (182/6106) other than Down syndrome	eligibility for social and educational services for ID	not given	register	British pediatric association ICD-9

Table 3: (continued).

Congenital malformation other chromosomal , endocrine / metabolic diseases					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Yeargin-Allsop et al. (1997)	1.3% (9/715) malformations of the nervous system[d] 2.0% (14/715) other malformations[e] 0.8% (6/715) metabolic, endocrine -/ thyroid gland disorders[f]	IQ test	DSM III	medical files	not given
Hou et al. (1998)	2.7% (322/11892) other malformations 0.2% (41/11892) metabolic, endocrine -/ thyroid gland disorders	family history/ medical records/ blood and urine tests	not given	clinical evaluation, blood sample	not given
Fernell (1998)	14.1% (9/64) malformations of the nervous system 7.8% (5/64) other malformations 1.6% (1/64) metabolic, endocrine -/ thyroid gland disorders	IQ and DQ tests	not given	medical files	not given
Cans et al. (1999)	2.2% (25/1150) malformations of the nervous system 6.4% (74/1150) other malformations 1.8% (21/1150) metabolic, endocrine -/ thyroid gland disorders	IQ test	ICD 10	medical files	not given
Airaksinen et al. (2000)	12.0% (18/151) malformations of the nervous system 11.3% (17/151) other malformations	school achievement / psychological tests or eligibility for social services for ID	ICD 10	clinical examination	not given
Stromme et al. (2000a)	7.3% (13/178) malformations of the nervous system 6.7% (12/178) other malformations 5.2% (4/178) metabolic, endocrine -/ thyroid gland disorders	IQ and DQ tests	DSM IV	medical files	not given
Christianson et al. (2002)	8.4% (20/238) malformations of the nervous system 0.8% (2/238) other malformations	IQ and DQ tests	not given	clinical examination	not given
Zhang (2005)	2.5% (2/79) other malformations	DQ tests	ICD 10	clinical examination	not given
Petterson et al. (2007)	5.6% (342/6106) malformations of the nervous system 13.1% (799/6106) other malformations	eligibility for social and educational services for ID	not given	register	British pediatric association ICD-9

[d] e.g. spina bifida, hydrocephaly
[e] e.g. Prader-Willi, tuberous sclerosis, Smith-Lemli-Opitz, malformations musculoskeletal system, genital/urinary digestive circulatory system
[f] e.g. phenylketonuria, hypothyroidism

Table 3: (continued).

Autistic disorder (AD)					
Study	Prevalence % (number/ respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Nordin et al. (1996)	8.9% (9/101) autistic disorder	IQ tests	not given	ABC, CARS, clinical examination	DSM III R
Fernell (1998)	17.2% (11/64) autism	IQ and DQ tests	not given	medical files	not given
Stromme et al. (2000b)	4.5% (8/178) childhood autism	IQ and DQ tests	DSM IV	clinical examination	ICD 10
Magnusson et al. (2001)	not given (32 /not given) childhood autism	IQ and DQ tests	not given	ADI-R, CARS, clinical examination	ICD 9 or 10
Molteno et al. (2001)	9.4% (12/128) autism	eligibility for special schools for ID	not given	DBC-T en CSI, clinical examination	not given
Koskentausta et al. (2002)	5.2% (8/155) childhood autism	IQ and DQ tests	ICD 10	medical files	ICD 10
de Bildt et al. (2005)	8.8 % (weighted score/825) autistic disorder	IQ and DQ tests	DSM IV TR	PDD-MRS, ADI-R, ADO-G, clinical examination	DSM IV TR
Zhang (2005)	10.1% (8/79) autistic disorder	DQ tests	ICD 10	CABS, CARS, clinical examination	DSM IV
Bradley et al. (2006a,b), Bryson et al. (2008)	25.1% (43/171) autism	IQ and DQ tests	not given	ADI-R, clinical examination	DSM IV

Pervasive developmental disorders (PDD) other than autistic disorder (AD)					
Study	Prevalence % (number/ respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Nordin et al. (1996)	3.0% (3/101) autistic-like disorder 7.9% (8/101) autistic disorder-NOS	IQ tests	not given	ABC, CARS, clinical examination	DSM III R
Fernell (1998)	6.3% (4/64) autistic symptoms	IQ and DQ tests	not given	medical files	not given
Airaksinen et al. (2000)	0.6% (1/151) Rett syndrome	school achievement / psychological tests or eligibility for social services for ID	ICD 10	clinical examination	not given

Stromme et al. (2000b)	0.6% (1/178) Rett syndrome 0.6% (1/178) Asperger 2.5% (4/178) PDD Unspecified	IQ and DQ tests	DSM IV	clinical examination	ICD 10
Magnusson et al. (2001)	not given (13 / not given) atypical autism	IQ and DQ tests	not given	ADI-R, CARS, clinical examination	ICD 9 or 10
Koskentausta et al. (2002)	7.7% (12/155) atypical autism	IQ and DQ tests	ICD 10	medical files	ICD 10
de Bildt et al. (2005)	7.9 % (weighted score/825) PDD-NOS	IQ and DQ tests	DSM IV TR	PDD-MRS, ADI-R, ADOS-G, clinical examination	DSM IV TR

ADHD / hyperkinetic disorder

Study	Prevalence % (number/ respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
Stromme et al. (2000b)	16.0% (28/178) hyperkinetic disorder	IQ and DQ tests	DSM IV	clinical examination	ICD 10
Molteno et al. (2001)	6.3% (8/128) ADHD	eligibility for special schools for ID	not given	DBC-T en CSI, clinical examination	not given
Koskentausta et al. (2002)	6.5% (10/155) hyperkinetic disorder	IQ and DQ tests	ICD 10	medical files	ICD 10
Dekker et al. (2003)	5.9% (28/474) ADHD	eligibility for special schools for ID	not given	DISC-IV-P, clinical examination	DSM IV
Emerson (2003),Emerson et al. (2007)	8.3% (53/641) hyperkinetic disorder	Primary carer reported ID and / or teacher reported ID	not given	DAWBA, clinical examination	ICD 10
Voigt et al. (2006)	30.0% (21/70) ADHD	IQ test	not given	ADHD questionnaires, clinical examination	DSM IV
Gothelf et al. (2008)	18.3% (16/87) ADHD	IQ test	not given	K-SADS-PL, clinical examination	DSM IV TR

Table 3: continued.

Miscellaneous psychiatric disorders					
Study	Prevalence % (number / respondents with ID)	Method of diagnosis ID	Classification framework ID	Method of diagnosis CD	Classification framework CD
van Schrojenstein Lantman et al. (1997)	2.0% (2/101) affective disorder 9.9% (10/101) other psychiatrics disorders	eligibility for residential centres for ID	not given	medical files	ICD 9
Stromme et al. (2000b)	3.0% (6/178) conduct disorder 6.0% (10/178) other behavioural/emotional disorder 3.0% (5/178) anxiety/phobic/obsessive-compulsive disorder 1.0 % (1/178) tic disorder	IQ and DQ tests	DSM IV	clinical examination	ICD 10
Koskentausta et al. (2002)	0.6 % (1/155) conduct disorder 0.6% (1/155) recurrent depressive disorder 1.3% (2/155) emotional disorder	IQ and DQ tests	ICD 10	medical files	ICD 10
Dekker et al. (2003)	3.0% (14/474) conduct disorder 13.9% (66/474) ODD 21.9% (103/474) any anxiety disorder 4.4% (21/474) any mood disorder	eligibility for special schools for ID	not given	DISC-IV-P, clinical examination	DSM IV
Emerson (2003),Emerson et al. (2007)	8.4% (54/641) conduct disorder 11.1% (71/641) ODD 1.4% (9/641) any depressive disorder 12.0% (77/641) any emotional disorder 11.4% (73/641)any anxiety disorder 0.8% (5/641) tic disorder 0.2% (1/641) eating disorder	Primary carer reported ID and / or teacher reported ID	not given	DAWBA, clinical examination	ICD 10

Gothelf et al. (2008)	4.6% (4/87) conduct disorder 13.8% (12/87) ODD 14.0% (12/87) any mood disorder 39.0% (30/87) any anxiety disorder 2.2% (2/87) any psychotic disorder 4.6% (4/87) tic disorder 3.4% (3/87) eating disorder 8.0% (7/87) impulse control disorder 4.6% (4/87) somatoform disorder 17.2% (15/87) enuresis/encopresis	IQ test	not given	K-SADS-PL, CY-BOCS, SBS, clinical examination	DSM IV TR

Abbreviations diseases: CD=Chronic diseases, COPD= Chronic Obstructive Pulmonary Disease, ID=Intellectual Disability, NOS= Not Otherwise Specified, ODD=Oppositional Defiant Disorder, PDD= Pervasive Developmental Disorder
Abbreviations classification framework: DSM= Diagnostic and Statistical Manual of Mental Disorders, IAPB= International Association for Prevention of Blindness , ILAE= International League Against Epilepsy, ICD = International Statistical Classification of Diseases.
Abbreviations method of diagnosis: ABC= Autism Behaviour Scale, ASQ= Autism Screening Questionnaire, ADI-R=Autism Diagnostic Interview-Revised, ADOS-G=Autism Diagnostic Observation Schedule-Generic, ASQ=Autism Screening Questionnaire, CABS= Clancy Autism Behaviour Scale, CARS= Children Autism Rating Scale, CSI= Child Symptom Inventory, CY-BOCS=Children's Yale-Brown Obsessive Compulsive Scale, DAWBA= Development and Well-Being Assessment, DBC-T= Developmental Behaviour Checklist-Teacher version, DISC-IV-P= Diagnostic Schedule for Children-Parent version, DQ= Development Quotient, IQ= Intelligence Quotient, K-SADS-PL=Kiddie Schedule for Affective Disorders and Schizophrenia for School Age Children Present and Lifetime Version, PDD-MRS= Pervasive Developmental Disorder in Mentally Retarded Scale, SBS=Stereotyped Behavior Scale, VABS=Vineland Adaptive Behaviour Scales.

2.3.3.2. Cerebral palsy (CP)

Prevalence rates of CP were reported in 11 studies and ranged from 8.4%-33.8%. The weighted mean prevalence rate was 19.8/100 (CI 18.6-21.1).

2.3.3.3. Visual problems

Four studies reported about visual problems such as refractive errors, strabismus, visual acuity, visual field or visual impairment in general. Prevalence rates ranged from 2.2%-26.8%. We were not able to calculate a weighted mean prevalence rate for the different visual problems because of the variability between the studies regarding outcomes.

Table 4: Prevalence rates range, weighted mean prevalence rates and 95% confidence (95% CI) interval of chronic diseases.

Chronic diseases	Prevalence rates (range)	Weighted mean prevalence rate	95% CI
Somatic			
Epilepsy	5.5%-35.0%	22.0/100 (943/4296)	(20.8;23.2)
Cerebral Palsy	8.4%-33.8%	19.8/100 (765/3868)	(18.6;21.1)
Hearing problems	0.0%-7.1%	4.5/100 (59/1320)	(3.4;5.7)
Cerebrovascular accident	0.3%-2.5%	2.0/100 (285/14014)	(1.8;2.3)
Congenital malformations			
Down syndrome	2.1%-20.3%	11.0/100 (2282/20817)	(10.5;11.4)
Fragile-X	0.1%-2.4%	1.9/100 (237/12785)	(1.6;2.1)
Cri-du Chat	0.1%-1.1%	0.2/100 (29/12785)	(0.2;0.3)
Other than Down Syndrome	0.3%-8.5%	3.1/100 (615/20041)	(2.9;3.3)
Malformation of the nervous system	1.3%-14.1%	5.1/100 (436/8602)	(4.6;5.6)
Other malformations	0.8%-13.1%	6.1/100 (1247/20573)	(5.7;6.4)
Metabolic or endocrine -/ thyroid gland disorders	0.2%-5.2%	0.5/100 (73/13999)	(0.4;0.7)
Mental			
Autistic disorder (AD)	4.5%-25.1%	10.1/100 (172/1701)	(8.8;11.6)
PDD categories that are not otherwise specified	2.5%-7.9%	7.1/100 (89/1259)	(5.8;8.6)
ADHD / hyperkinetic disorder	5.9%-30.0 %	9.5/100 (164/1733)	(8.2;10.9)
Conduct disorder	0.6%-8.4%	5.1/100 (79/1535)	(4.1;6.4)
Oppositional Defiant Disorder	11.1%-13.9%	12.4/100 (149/1202)	(10.7;14.4)
Any anxiety disorder	11.4%-39.0%	17.1/100 (206/1202)	(15.1;19.4)
Tic disorder	0.8%-4.6%	1.1/100 (10/906)	(0.6;2.0)

2.3.3.4. Hearing problems

Five studies reported about hearing impairment or disability in general. Prevalence rates ranged from 0.0%-7.1%. The weighted mean prevalence rate was 4.5/100 (CI 3.4-5.7).

2.3.3.5. Miscellaneous somatic chronic diseases

Seven studies reported prevalence rates of miscellaneous somatic chronic diseases such as COPD (8.9%), gastric & oesophageal diseases (6.9%), back & neck disorders (6.9%), osteoarthropathia (2.5%), cerebrovascular accident (CVA) (range: 0.3%-2.5%), Reye syndrome (0.3%), human immunodeficiency virus (0.0%) and other chronic diseases (4%). The weighted mean prevalence rate for CVA was 2.0/100 (CI 1.8-2.3). For the other chronic diseases we were not able to calculate a weighted mean prevalence rate.

2.3.3.6. Congenital malformations genetic-chromosomal and sex-linked

Ten studies reported about congenital chromosomal malformation. Prevalence rates of Down syndrome, fragile-X, and Cri-du-Chat and other than Down syndrome chromosomal malformation (included fragile-X, Cri-du-Chat, etc.) were reported and ranged from 0.1-20.3% for the different disorders. The weighted mean prevalence rate for Down syndrome was 11.0/100 (CI 10.5-11.4), for fragile-X 1.9/100 (CI 1.6-2.1), for Cri-du-Chat 0.2/100 (CI 0.2-0.3) and for other than Down syndrome 3.1/100 (CI 2.9-3.3).

2.3.3.7. Congenital malformations other chromosomal, endocrine / metabolic diseases

Nine studies reported prevalence rates of congenital malformations other chromosomal, endocrine / metabolic diseases. Prevalence rates of malformations of the nervous system (e.g. spina bifida, hydrocephaly), other malformations (e.g. Prader-Willi, tuberous sclerosis, malformations of the musculoskeletal -, genital/ urinary -, digestive - or circulatory system) and metabolic-, endocrine -/ thyroid gland disorders (e.g. phenylketonuria, hypothyroidism) were reported and ranged from 0.8%-13.1% for the different disorders. The weighted mean prevalence rate for malformations of the nervous system was 5.1/100 (CI 4.6-5.6), for other malformations 6.1/100 (CI 5.7-6.4) and for metabolic -, endocrine -/ thyroid gland disorders 0.5/100 (CI 0.4-0.7).

2.3.3.8. Autistic disorder (AD)

Prevalence rates of AD were reported in 11 studies and ranged from 4.5%-25.1%. The weighted mean prevalence rate was 10.1/100 (CI 8.8-11.6).

2.3.3.9. Pervasive developmental disorders (PDD) other than autistic disorder (AD)

Prevalence rates of PDD other than AD, including Rett syndrome, Asperger syndrome, atypical autism, PDD not otherwise specified (NOS), autistic symptoms, autistic like disorder and autistic disorder-NOS, were reported in seven studies and ranged from 0.6%-7.9% for the different disorders. Prevalence rates of the PDD categories that are not otherwise specified, such as PDD-NOS, atypical autism and autistic disorder-NOS were reported in four studies and ranged from 2.5%-7.9%. The weighted mean prevalence rate for PDD categories that are not otherwise specified was 7.1/100 (CI 5.8-8.6). For the other diseases we were not able to calculate a weighted mean prevalence rate.

2.3.3.10. ADHD / hyperkinetic disorder

Prevalence rates of ADHD / hyperkinetic disorder were reported in eight studies and ranged from 5.9%-30.0 %. The weighted mean prevalence rate was 9.5/100 (CI 8.2-10.9).

2.3.3.11. Miscellaneous psychiatric disorders

Seven studies reported prevalence rates of miscellaneous psychiatric disorders. Most frequently mentioned disorders are: conduct disorder with prevalence rates ranged from 0.6%-8.4%; oppositional defiant disorder (ODD) with prevalence rates ranged from 11.1%-13.9%; any anxiety disorder with prevalence rates ranged from 11.4%-39.0%; and tic disorder with prevalence rates ranged from 0.8%-4.6%. The weighted mean prevalence rate for conduct disorder was 5.1/100 (CI 4.1-6.4), for oppositional defiant disorder 12.4/100 (CI 10.7-14.4), for any anxiety disorder 17.1/100 (CI 15.1-19.4), and for tic disorder 1.1/100 (CI 0.6-2.0). Prevalence rates of enuresis/encopresis, affective -, behavioural/emotional -, anxiety/phobic/ obsessive-compulsive -, depressive -, emotional -, mood -, psychotic -, eating - , impulse control -, and somatoform disorders were also reported and ranged from 0.6%-17.2% for the different disorders. We were not able to calculate a weighted mean prevalence rate for these disorders.

2.4 Discussion

This systematic literature review shows high prevalence rates of a wide range of chronic diseases in children with ID. Thirty-one studies that focused on a limited amount of chronic diseases had a sufficient methodological quality and were included. The quality of these studies was in general good to high. However, the prevalence rates for most chronic diseases varied between the included studies. The characteristics of the sample, the recruitment of the study population, the method of diagnosis, the classification framework that was used and factors that were not examined and appraised in this study like the quality of registers, diagnostic overshadowing, or the accessibility of healthcare for children with ID in different countries, can contribute to this variation [6-8, 64, 65]. Nevertheless, the prevalence rates of chronic diseases in children with ID are higher than the prevalence rates of chronic diseases reported in studies on children without ID [66-76]. It should be noted that in contrast to the reviews of Fombonne [69, 71, 77], we found higher prevalence rates of autistic disorder compared to pervasive developmental disorders other than autistic disorder. An explanation could be the inclusion of studies reporting about children without ID in Fombonne's reviews. Children with autistic disorder are more likely to have ID than children with PDD other than autistic disorder [78]. However, Bertrand et al. [79] and Williams et al. [80] also reported higher prevalence rates of autistic disorder compared to PDD-NOS in population studies of children with and without ID.

2.4.1. Fit with previous studies

To our knowledge this is the first systematic review on chronic diseases in children with ID. Comparison of our results with other reviews on chronic diseases in children with ID is difficult [4, 28, 68, 81-85]. In contrast to traditional narrative reviews, we appraised the studies on methodological characteristics reflecting the internal and external validity.

2.4.2. Strengths and limitations

A major strength of our study is the very thorough search strategy that covered all relevant literature databases and included a check of the references of the papers that were found. Despite this, we may still have missed some publications, e.g. studies that are not indexed well in the databases. Another strength of our study is that we used a consensus on chronic diseases and health conditions to define chronic disease in childhood [25], which is operationalised in a list of chronic diseases by using the ICD [17, 18] and the DSM [19-22].

The generalization of the results is limited to the chronic diseases reported in the studies we included in the systematic review and to the so-called in particular European "industrialized countries". We hardly found studies from the so-called "less industrialized countries". Another limitation was the focus on studies reporting about chronic diseases in children with ID in general. Excluding studies that were focused exclusively on subpopulations of children with a specific biomedical cause of ID such as Down syndrome does not appreciate the prevalence of syndrome-related health problems associated with this condition. Diseases such as congenital heart disorders, obesity, celiac disease, thyroid disorders are very prevalent in children with Down syndrome, but were not or hardly reported in studies on children with ID in general. Inclusion of studies on children with Down syndrome would increase the prevalence of these conditions in the population with ID. However, reviews about the prevalence of chronic diseases in persons with Down syndrome are already available [26-28].

2.4.3. Implications for clinicians

The height of the prevalence rates should alert clinicians to the widespread of chronic diseases in children with ID. Clinicians are crucial in the identification and registration of chronic diseases. Identification begins with a complete history, including a pre-, peri- and postnatal medical history, physical examination and psychological and social evaluation [4, 10, 11, 86]. Early detection and adequate treatment of chronic diseases is important because these conditions have a significant negative impact on the well-being and social participation of children with ID and their families [2, 10-16]. Moreover, a good diagnostic work-up according to professional standards accompanied with a good registration system are useful tools to delineate (probable) causes [14]. Evidence on the association between ID and chronic diseases may identify etiologic clues that are necessary for the early identification of these conditions and the development and implementation of effective programs to increase the opportunities for children with ID [10, 40, 61, 87].

2.4.4. Implications for research

To improve evidence on the prevalence of chronic diseases in children with ID researchers have to reach consensus about the classification framework that should be used in prevalence studies for psychiatric disorders in children with ID in the first place. A promising development is the future alignment of the DSM-V with the ICD-11. This will present an opportunity to unify and strengthen knowledge of global mental health. However, the debate about the validity of both classification systems as a clinical and research tool is not finished yet [88-91]. Other developments

in this direction that could be used are the Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation developed by the Royal College of Psychiatrists [92, 93], or the Developmental Psychiatric Assessment as developed by Dosen [94, 95]. Second, the use of multiple data sources, in contrast to the one data source strategy used in most studies, will improve the validity of the prevalence rates [16]. Third, transnational comparative population studies are needed including also the so-called "less industrialized countries". Fourth, reporting of the studies should be improved to enable a proper assessment of the internal and external validity of the included studies. This may be reached by using the recently developed STROBE guidelines [30].

Our review shows that much can be gained in the caring for children with ID by better evidence on the occurrence of chronic diseases.

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Chapter 3

Prevalence of chronic diseases in adolescents with intellectual disability

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Abstract

Valid community-based data on the prevalence of chronic diseases in adolescents (12-18 years) with intellectual disability (ID-adolescents) are scarce. The aim of this study is to assess the prevalence rates and the nature of chronic diseases in a population of ID-adolescents and to compare them with rates among adolescents in the general population. Therefore, we obtained data on 1083 ID-adolescents attending secondary schools, day care centres or living in residential centres fully covering one region of the Netherlands. Parents of the adolescents completed a questionnaire about the occurrence of chronic diseases in their child during the previous twelve months and about background characteristics. The questionnaire was derived from the Dutch National Permanent Survey on Living Conditions questionnaire periodically administered in a representative population sample (n≈10.000). Prevalence rates of chronic diseases in ID-adolescents were compared with those in adolescents in the Dutch general population. Among ID adolescents, high prevalence rates of a wide range of chronic diseases were found. The five most prevalent were: ADHD (21.1%), PDD-NOS (14.0%), dyslexia (13.9%), migraine or chronic headache (12.7%), and autistic disorder (10.9%). These prevalence rates were all higher ($p < 0.05$) than among adolescents in the general population. Of all ID-adolescents, 62.9% was reported to have at least one chronic disease. The burden of chronic diseases among ID-adolescents is very high, showing a high need for adequate care. These high prevalence rates should alert policymakers and clinicians regarding the widespread of chronic diseases among ID-adolescents.

3.1. Introduction

Valid community-based data on the prevalence rates of the full range of chronic diseases in adolescents (12-18 years) with intellectual disability (ID-adolescents) are scarce [1, 2]. A number of studies have reported on the prevalence of specific chronic diseases in young people with ID [3-33]. However, only few studies have reported such prevalence rates of a wide range of chronic diseases associated with ID [13, 28-30]. Moreover, only some studies compared their results with the prevalence rates of chronic diseases in the general population [11, 12, 19, 26, 31], and none of these studies reported prevalence rates of chronic diseases among ID-adolescents.

Adolescence is a specific stage of life between child- and adulthood with specific health needs. It is a time of life marked by physical, emotional, behavioural and social changes, but also by relatively high risks for the onset of (chronic) health problems [1, 34, 35]. Literature about young people with ID suggests that adolescents have a greater risk on chronic diseases compared to adolescents without ID, but inclusive data on this are lacking [2, 11, 12, 19, 26, 31, 36-38].

Age-specific community-based data are thus needed to support policymakers and professionals in the adequate provision and planning of care to ID-adolescents. Policymakers need these data for the planning and financing of adequate care arrangements (e.g. health, education, work) to enhance the well being and societal participation of ID-adolescents and their families. Professionals need these data to know who are at risk for chronic diseases and to prevent chronic diseases, to support the early detection and adequate treatment of chronic diseases and their consequences among ID-adolescents and their families [39-42].

The aim of this study is: (1) to assess the prevalence and the nature of chronic diseases in a population of ID-adolescents; (2) to compare the prevalence rates of chronic diseases in ID-adolescents with that among adolescents in the general population.

3.2. Methods

3.2.1. Participants

3.2.1.1. Adolescents with ID

We collected data in 2006-2007 from adolescents with a borderline, mild, moderate, severe or profound ID aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people). Adolescents of the target population attended secondary schools, special

secondary schools, day care centres or lived in residential centres. ID-adolescents attending secondary schools (schools for practical training) can be classified as mainly educable and have IQs between 60 and 84. ID-adolescents attending special secondary schools can be classified as mainly trainable and have IQs between 30 and 59. ID-adolescents not attending school, most of them with IQs < 30, attend day care centres or live in residential centres [43]. The target population had been officially classified as having ID by two independent committees. The Dutch Ministry of Education, Culture and Science established a committee for adolescents attending secondary or special secondary schools and the Dutch Ministry of Health, Welfare and Sport established a committee for adolescents attending day care centres or those living in residential centres. The classification of ID is based on information from validated intelligence tests obtained by both committees [44, 45].

In the current community-based cross-sectional research project, 70% of the schools and centres in both the provinces participated with a total of 2271 adolescents. Non-participating schools and centres did not differ from participating schools and centres with regard to urbanisation of the catchment area and number of adolescents. All parents of the 2271 adolescents aged 12-18 years received a questionnaire and a reminder when they did not respond; 1083 parents returned the questionnaire (48%). Adolescents in the response and non-response group did not differ with regard to age (t-test=1.86, ns), but in the response group girls ($\chi 2 = 4.35$; $p < 0.05$) and adolescents with borderline or mild ID ($\chi 2 = 8.96$; $p < 0.05$) were somewhat overrepresented. However, the effect sizes for both variables were negligible; Cohen's *w* were 0.05 and 0.07, respectively [46].

The study protocol was approved by the Medical Ethics Committee of the University Medical Centre Groningen, the Netherlands.

3.2.1.2. Adolescents in the general population

Statistics Netherlands conducts yearly the National Permanent Survey on Living Conditions in a representative sample (n≈10.000) of the Dutch population [47]. We used the data, adjusted for non-response, on adolescents aged 12-18 years in 2007 (response: 64%)[48]. Prevalence data on chronic diseases in adolescents in the general population that were not available via Statistics Netherlands were derived from the National Institute for Public Health and the Environment (NIPHE). The NIPHE provides national and international data to support policymakers and professionals in various fields of work such as prevalence rates of chronic diseases [49].

3.2.2. Measures

3.2.2.1. Chronic diseases in ID-adolescents

Chronic diseases in ID-adolescents were measured by the National Permanent Survey on Living Conditions (POLS) questionnaire; module health and labour, part chronic diseases in children [50]. POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic systems and attention deficit hyperactivity disorder (ADHD). Questions were added about the presence of pervasive developmental disorders (PDD). Parents were asked to fill in the presence or absence of each specific chronic disease in the last 12 months for their child. Parents were also offered the possibility to mention chronic diseases that were not listed in the questionnaire.

3.2.2.2. Chronic diseases in adolescents in the general population

The prevalence rates of chronic diseases in adolescents in the general population were measured in the same way as in our study, using the POLS questionnaire. Prevalence rates of chronic diseases in adolescents in the general population that were not available in POLS were obtained from the NIPHE. The NIPHE data are based on national and international research [49].

3.2.2.3. Background characteristics of ID-adolescents

The questionnaire in the ID-adolescents sample comprised questions on age, gender and type of school or institution the adolescent is attending. We used type of school or institution as a proxy for severity of ID.

3.2.3. Analysis

First, we computed prevalence rates and the 95% confidence intervals (95% CI) of having one or more chronic diseases, overall and by type of chronic diseases. The association between the number of chronic diseases and ID severity and gender were assessed via Univariate analysis of variance parameter estimates. Second, we performed multinomial logistic regression analyses. Odds ratios (OR) and their 95% confidence interval (95% CI) were computed to assess the association between the occurrence of chronic diseases by type, and ID severity and gender. Third, we computed prevalence rates of separate chronic diseases in ID-adolescents, and compared these rates with rates among adolescents in the general population (12-18 years) [48]. We tested differences using chi-square tests. If national data were not available from Statistics Netherlands, we used data from

the NIPHE instead [49]. Differences were tested via single proportion tests. For all the differences in proportions, effect sizes according to Cohen's *h* were calculated [46]. Cohen (1988) defines an effect sizes of <0.20 as trivial effects; of ≥0.20 to <0.50 as small effects; of ≥0.50 to <0.80 as moderate effects; and of ≥0.80 as large effects.

3. 3. Results

Table 1 shows the background characteristics of the adolescents. The gender ratio, 58.3% boys and 41.7% girls, was similar to the ratio boys and girls with ID in the Netherlands [51].

Table 1: Demographic characteristics of the sample

Characteristics	Years
Age (n=1066)	
Mean (SD)	15.4 (1.6)
Range	12-18
Gender (n=1074)	n (%)
Boys	626 (58.3)
Girls	448 (41.7)
Level of ID[a] (n=1077)	n (%)
IQ 60-80	785 (72.9)
IQ 30-59	253 (23.5)
IQ < 30	39 (3.6)

[a] derived from the type of institution at which they were allocated

Table 2 shows that 63% of the ID-adolescents had at least one chronic disease and 34% had two or more chronic diseases (mean 1.30; SD 1.47). Thirty-nine per cent of the ID-adolescents had one or more somatic chronic diseases and 41% one or more mental chronic diseases. Out of these, 20% had a combination of somatic and mental chronic diseases.

Table 2: Number and type of chronic disease(s) and 95% confidence (95% CI) intervals in ID-adolescents

	ID-adolescents (n=1083)		
Number of chronic diseases	n	%	95% CI
0	402	37.1	(34.29;40.04)
1	313	28.9	(26.28;31.67)
2	178	16.4	(14.35;18.77)
3	97	9.0	(7.40;10.81)
4	56	5.2	(4.00;6.65)
5 -11	37	3.4	(2.49;4.68)
Type of chronic diseases			
None	402	37.1	(34.29;40.04)
Somatic chronic disease(s)	235	21.7	(19.35;24.25)
Mental chronic disease(s)	245	22.6	(20.23;25.21)
Somatic & mental chronic diseases in combination	201	18.6	(16.36;20.99)

Gender and severity of ID were associated with these prevalence rates. Gender was not associated with the number of chronic diseases, but boys were more likely than girls to be diagnosed with mental chronic diseases (OR 2.23; 95%CI 1.57-3.16) and with a combination of somatic and mental chronic diseases (OR 1.47; 95%CI 1.03-2.10). Severity of ID was positively associated with the number of chronic diseases ($F=48.12$, $p < 0.000$). With regard to the type of the chronic diseases, adolescents with severe and moderate ID were more likely than adolescents with mild ID to be diagnosed with somatic chronic diseases (OR (95%CI) 12.35 (2.70-55.56), and 3.31 (2.04-5.00), respectively), mental chronic diseases (9.00 (1.91-41.67), and 1.88 (1.22-2.86), respectively) and a combination of somatic and mental chronic diseases (25.00 (5.62-111.11), and 3.85 (2.56-5.88), respectively). In addition, adolescents with severe ID were more likely than adolescents with moderate ID to be diagnosed with a combination of somatic and mental chronic diseases (6.44 (1.43-29.14)).

Table 3 shows that the prevalence rates of chronic diseases among ID-adolescents were statistically significant higher for 8 of the 17 chronic diseases compared to the prevalence rates of these chronic diseases in adolescents in the general population. The effect sizes were negligible for two (asthma, chronic bronchitis and COPD, and psoriasis) of the four somatic chronic diseases that differed with statistical significance, and small for the two other ones (epilepsy, heart and blood diseases). With regard to mental chronic diseases, the effect sizes for the statistically significant differences were small for dyslexia and medium for ADHD, PDD-NOS and autistic disorder.

Table 3: Prevalence rates of chronic diseases in adolescents with ID and in the general population

	ID-adolescents		General population		Differences	
Somatic chronic diseases	%	n/N	%	n/N	p-Value	Cohen's h[c]
Asthma, chronic bronchitis, COPD[a]	9.9	107/1083	6.2	30/481	**0.019**	0.13
Chronic eczema[a]	4.3	47/1083	2.6	14/536	n.s.[d]	0.09
Diabetes[a]	0.5	5/1083	0.4	2/498	n.s.	0.01
Gastrointestinal and liver diseases[a]	1.8	19/1083	1.0	4/396	n.s.	0.06
Migraine or chronic headache[a]	12.7	138/1083	11.5	52/452	n.s.	0.04
Musculoskeletal diseases[a]						
Diseases of the back[a]	2.6	28/1083	2.7	14/517	n.s.	0.01
Inflammatory polyarthropathies[a]	0.8	9/1083	0.4	2/442	n.s.	0.05
Diseases of neck, shoulder and upper extremities[a]	5.3	57/1083	3.3	14/430	n.s.	0.10
Psoriasis[a]	1.3	14/1083	0.1	1/519	**0.030**	0.14
Heart - and blood diseases[a]	2.4	26/1083	0.1	1/524	**0.001**	0.22
Congenital malformations circulatory system[b]	2.1	23/1083	0.62[e]	-	n.s.	0.13
Congenital malformations nervous system[b]	2.0	22/1083	0.21[e]	-	n.s.	0.19
Epilepsy[b]	5.3	57/1083	0.27[f]	-	**0.000**	0.36
Somatic chronic diseases no reference data available			95% CI			
Congenital malformations eye	3.0	32/1083	(2.10;4.14)			
Congenital malformations ear	1.8	20/1083	(1.20;2.84)			
Chromosome abnormalities	3.5	38/1083	(2.57;4.78)			
Cerebral Palsy	0.5	5/1083	(0.20-1.07)			
Muscular diseases	0.7	8/1083	(0.38-1.45)			
Other congenital malformations	6.1	66/1083	(4.81;7.68)			
Other somatic disorders	3.7	40/1083	(2.72-4.99)			

Mental chronic diseases	%	n/N	%	n/N	p-Value	Cohen's h[c]
Dyslexia[a]	13.9	151/1083	3.8[g]	87/2285	**0.000**	0.37
Attention deficit/hyperactivity disorder (ADHD)[b]	21.1	229/1083	1.3[h]	-	**0.000**	0.73
Autistic disorder[b]	10.9	118/1083	0.1[i]	-	**0.000**	0.61
Pervasive developmental disorder not otherwise specified (PDD-NOS)[b]	14.0	152/1083	0.2[i]	-	**0.000**	0.68
Mental chronic diseases no reference data available			95% CI			
Other psychiatric disorders	1.8	19/1083	(1.12;2.72)			

Bold results are statistically significant.
[a] Reference data on adolescents aged 12-18 from Statistics Netherlands (POLS)
[b] Reference data from the National Institute for Public Health and the Environment
[c] Cohen thresholds: negligible effect (≤0.20); small effect (≥0.20 and <0.50); medium effect (≥0.50 and <0.80); large effect (≥0.80)
[d] Not significant
[e] Only data at birth available.
[f] Only data on the age-group 15-24 years available
[g] Only data on the age-group 4-12 years available
[h] Only data on the age-group 13-17 years available
[i] Prevalence data estimated on the basis of the review of Fombonne[52] on autistic disorder and other pervasive developmental disorders

3.4. Discussion

This study shows high prevalence rates of a wide range of chronic diseases in ID-adolescents. For 8 of the 17 chronic diseases that we assessed the prevalence rates were statistically significant higher among ID-adolescents than among in adolescents in the general population. Differences were particularly large with regard to ADHD, autistic disorder, and PDD-NOS, and smaller for some somatic diseases. Moreover, differences in prevalence rates were larger if ID was more severe, with regard to any somatic chronic diseases, any mental chronic diseases and with regard to a combination of somatic and any mental chronic diseases.

3.4.1. Fit with previous studies

Our findings regarding ID-adolescents cannot be compared with previous ones on adolescents because our study was the first that examined the prevalence rates of a wide range of chronic diseases in ID-adolescents, and compared them with the prevalence rates among adolescents in the general population. Previous studies on youth with ID also found higher prevalence rates of a wide range of mental chronic diseases [11, 12, 19, 31] and a wide range of congenital malformations [26] though. However, these studies did not separately report on adolescents, thus failing to recognize adolescence as a specific developmental stage. It is likely that this explain the differences in the prevalence rates of mental chronic diseases and congenital malformations these studies found compared to our study.

Three factors have been proposed to explain the high rates of chronic diseases in the ID population in general. First factor is biological / genetic, i.e. genetic and chromosomal disorders that cause both ID and a wide range of chronic somatic and mental diseases [11, 17, 53]. Second, the association of ID with socio-economic disadvantage may lead to adverse health outcomes and to higher rates of chronic diseases [11, 36, 54, 55]. Third, mental chronic diseases may be associated with ID because they share diagnostic characteristics [11, 53].

3.4.2 Strengths and limitations

Important strengths of this study are that it examined the prevalence rates of a wide range of chronic diseases in ID-adolescents in a community-based sample representative for about 90% of the adolescents with ID, and compared most outcomes with similarly obtained data on adolescents without ID. A limitation may be that data collection procedures may have slightly differed between adolescents with ID and in the general population. POLS did not cover mental disorders other than ADHD, we used data of the NIPHE instead. However, differences in prevalence

rates were that large that they are unlikely to be fully explained by methodological differences. A second limitation is the relatively low response rate of our study (48%). This could lead to selection bias, but non-response analyses revealed no major differences with regard to age, gender and educational level.

3.4.3 Implications

We found disconcertingly high prevalence rates for some chronic diseases among ID-adolescents which should alert policymakers and clinicians to these diseases among ID-adolescents. This shows a need for effective care arrangements to handle this huge burden of morbidity, both with regard to prevention, and treatment. As such, it provides a challenge to both clinicians and policy [39-42].

Our study is the first to examine the prevalence rates of a wide range of chronic diseases in ID-adolescents and compare the results with data on adolescents without ID. Therefore our findings need confirmation, including an assessment of the pathways leading to such high prevalence rates. Anyhow, our results show a very high burden of chronic diseases among ID-adolescents, and thus a high need for adequate care [2, 36] .

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Chapter 4

Limited concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents

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Abstract

Evidence on teachers' knowledge about somatic and mental chronic diseases among adolescents with intellectual disability (ID-adolescents) compared to the knowledge parents and healthcare professionals have, is limited. The aim of this study is: (1) to assess the knowledge of teachers on the presence of chronic diseases in ID-adolescents; (2) to compare teachers with parents and healthcare professionals and parents with healthcare professionals regarding the knowledge on the presence of chronic diseases in ID-adolescents. We obtained data on 1044 ID-adolescents attending secondary schools, fully covering one region of the Netherlands. Teachers, parents and general practitioners (GPs) of the adolescents completed a questionnaire about the occurrence of chronic diseases in their child, pupil, or patient, during the previous 12 months. The questionnaire was derived from the Dutch National Permanent Survey on Living Conditions questionnaire periodically administered in a representative population sample (n≈10.000). Concordance between teachers, parents and GPs on the presence of chronic diseases in ID-adolescents was relatively low. In about half of all 66 dyads the concordance was for the most part fair and just in 10 dyads good to very good; nine of these latter cases concerned somatic chronic diseases. In addition, teachers reported mostly lower prevalence rates of chronic diseases in ID-adolescents compared to the parents, in particular on mental chronic diseases. Although prevalence rates of chronic diseases among ID-adolescents are very high, knowledge on this among teachers is limited. While information on chronic diseases in ID-adolescents is available among different informants, the disagreement between them reflects different points of view between the informants and probably indicates a lack of communication. The communication among teachers, parents and GPs should be improved to combine the knowledge and information on the presence of chronic diseases in ID-adolescents. This may provide opportunities to improve the support of these adolescents in their school career and in their transition from school to work.

4.1. Introduction

Chronic diseases are more prevalent among adolescents with intellectual disability (ID-adolescents) than among adolescents without ID [1-6]. Several studies have indicated that chronic diseases in ID-adolescents, in particular mental chronic diseases, largely increase the likelihood of emotional and behavioural problems [7-13]. Both, emotional and behavioural problems and disease specific problems, such as pain, fatigue or deficits in attention or hyperactivity, have a profound effect on participation of ID-adolescents in educational programs, occupational opportunities, and also on the potential to live in the community [1, 4, 13-17].

Adequate school support highly contributes to the educational achievements of adolescents and their subsequent transition to employment [17-23]. Teachers play a pivotal role in the challenging process to prepare adolescent successfully for the workforce. In case of ID-adolescents, teachers are more challenged since most ID-adolescents have chronic diseases which affect educational success [18, 19, 23, 24]. Therefore, information on chronic diseases among ID-adolescents is highly needed by teachers, since it may help teachers to manage the effects of chronic diseases on the adolescents' emotional and behavioural functioning and their disabilities.

The limited evidence as available shows that teachers' knowledge about chronic diseases of their ID-adolescent students is much less than needed. This may be due to the fact that parents and healthcare professionals do not share this information with teachers [18, 19, 23, 25]. Diagnostic overshadowing among teachers may be the result, i.e. teachers attribute the adolescents' problem behaviour and disabilities to their ID rather than to other, potentially manageable causes related to their chronic disease(s) [26]. This misattribution may lead to the maintenance or aggravation of problem behaviour because effective interventions to prevent symptoms of the disease that elicit problem behaviour will not be used.

The aim of this study is therefore: (1) to assess the knowledge of teachers on the presence of chronic diseases in ID-adolescents; (2) to compare teachers with parents and healthcare professionals and parents with healthcare professionals regarding this knowledge on the presence of chronic diseases in ID-adolescents.

4.2. Methods

4.2.1. Participants

4.2.1.1. Sample

We collected data in 2006-2007 from adolescents with a borderline, mild, moderate or severe ID aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people). Nearly all adolescents of the target population attended secondary schools (schools for practical training) or special secondary schools (regional expertise centres). ID-adolescents attending schools for practical training can be classified as mainly educable and have IQs between 60 and 80. ID-adolescents attending regional expertise centres can be classified as mainly trainable and have IQs between 30 and 59 [27]. ID-adolescents not attending secondary schools, most of them with profound ID, were not included. The target population had been officially classified as having ID by an independent committee established by The Dutch Ministry of Education, Culture and Science. The classification of ID is based on information from validated intelligence tests [28].

In the current school-based cross-sectional study, 88% of the schools for practical training and regional expertise centres in both provinces participated. Non-participating schools did not differ from participating schools regarding urbanization of the catchment area and number of students. All parents of the 2156 adolescents aged 12-18 years received a questionnaire and a reminder when they did not respond. One thousand forty four parents (48%) returned the questionnaire. Adolescents in the response and non-response group did not differ regarding age (t-test = 1.751, ns), but the response group had a higher proportion of girls ($x2 = 5.9$; $p < 0.05$) and a higher proportion of adolescents with borderline or mild ID ($x2 = 9.8$; $p < 0.05$). However, the effect sizes for both variables were trivial; Cohen's *w* were 0.06 and 0.07, respectively [29].

4.2.1.2. Sub sample with data from parents, teachers and general practitioners

Additionally, parents were asked for informed consent to derive information on chronic diseases of their child from their teacher and healthcare professionals, in this case general practitioners (GPs). GPs are in the Netherlands the best setting to provide valid information on common and lifelong health problems [30]. Eight hundred ninety one (85%) parents gave informed consent. Teachers and GPs returned the questionnaires of 767 (86%) and 724 (81%) adolescents, respectively. Data on the presence of chronic diseases in ID-adolescents from all three informants were available for 539 adolescents. These 539 adolescents did

not differ regarding gender ($\chi 2 = 0.4$; ns) and IQ ($\chi 2 = 3.4$; ns), but they were older (t-test=2.2, $p < 0.05$) compared to the non-response group within this sub sample. However, the effect size was trivial; Cohen's *d* was 0.12 [29].
The study protocol was approved by the Medical Ethics Committee of the University Medical Centre Groningen, the Netherlands.

4.2.2. Measures

4.2.2.1. Chronic diseases in ID-adolescents

Chronic diseases in ID-adolescents were measured by the National Permanent Survey on Living Conditions questionnaire (POLS); module health and labour, part chronic diseases in children [31]. POLS was developed by Statistics Netherlands and is yearly used in a representative sample ($n \approx 10.000$) of the Dutch population [32]. POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic systems and ADHD. Questions were added about the presence of pervasive developmental disorders (PDD). Parents, and if informed consent was obtained from the parents, also teachers and GPs were asked to report the presence or absence of each specific chronic disease in the last 12 months for their child, pupil, or patient, respectively. They could also report the presence of chronic diseases that were not listed in the questionnaire.

4.2.2.2. Background characteristics

The questionnaire comprised questions on age, gender and school type of the adolescent. School type was used as proxy for severity of ID.

4.2.3. Analysis

First, we computed prevalence rates of the separate chronic diseases, based on teachers', parents' and GPs' report in 539 ID-adolescents. Second, we compared the prevalence rates teachers reported on the separate chronic diseases in ID-adolescents with that of the parents and GPs, respectively. In addition, the prevalence rates parents and GPs reported on the separate chronic diseases in ID-adolescents were compared. Differences were tested using chi-square tests and for all differences effect sizes according to Cohen, Cohen's *h*, were calculated [29]. Cohen [29] defines an effect size of <0.20 as trivial; of ≥0.20 to <0.50 as small; of ≥0.50 to <0.80 as moderate; and of ≥0.80 as large effect. Finally, the concordance between the knowledge of the three informants on the presence of chronic diseases in ID-adolescents was measured using Cohen's kappa for three

dyads: teacher-parent, teacher-GP and parent-GP. We used the guidelines for interpretation of kappa proposed by Landis and Koch [33]. Landis and Koch [33] defined a kappa of <0.21 as poor agreement; of ≥0.21 to <0.41 as fair agreement; of ≥0.41 to <0.61 as moderate agreement; of ≥0.61 to <0.81 as good agreement; and of ≥0.81 to 1.00 as very good agreement.

4.3. Results

Table 1 shows the background characteristics of the adolescents. The gender ratio, 59.3% boys and 40.7% girls, was similar to the ratio boys and girls with ID in the Netherlands [34].

Table 1: Demographic characteristics of the sample

Characteristics	
Age (n=539)	Years
Mean	15.7
SD	1.7
Range	12-18
Gender (n=538)	n (%)
Boys	319 (59.3)
Girls	219 (40.7)
Level of ID[a] (n=539)	n (%)
IQ 60–80	416 (77.2)
IQ 30–59	123 (22.8)

[a]as measured by school type

4.3.1. Differences between the informants

Table 2 shows that teachers reported statistically significant lower prevalence rates for eight chronic diseases compared to the adolescents' parent. The effect sizes were trivial for four chronic diseases (asthma, chronic bronchitis and COPD, chronic eczema, diseases of neck, shoulder and upper extremities, ADHD), and small for the four other ones (migraine or chronic headache, psoriasis, autistic disorder, dyslexia). Moreover, teachers reported statistically significant higher prevalence rates for two chronic diseases (migraine or chronic headache, dyslexia) compared to the GPs. Both effect sizes were small. Teachers reported statistically significant lower prevalence rates for two chronic diseases compared to the GPs. The effect size was small for asthma, chronic bronchitis, COPD and trivial for congenital malformations nervous system.

Table 2: Prevalence rates teachers, parents and GPs reported on the separate chronic diseases in ID-adolescents (n=539) and the differences between the three informants.

	Informants						Differences					
	Teachers		Parents		GPs		Teachers-Parents		Teachers-GPs		Parents-GPs	
Somatic chronic diseases	%	n	%	n	%	n	p-Value	h[a]	p-Value	h	p-Value	h
Asthma, chronic bronchitis, COPD	4.3	23	9.3	50	8.9	48	**0.001**[b]	0.20	**0.002**	0.19	0.832	0.01
Cerebral palsy	0.6	3	0.7	4	0.6	3	0.704	0.02	1.000	0.00	0.704	0.02
Chronic eczema	1.7	9	4.8	26	2.4	13	**0.003**	0.18	0.389	0.05	**0.033**	0.13
Congenital malformations circulatory system	0.9	5	1.9	10	2.2	12	0.194	0.08	0.087	0.11	0.667	0.03
Congenital malformations nervous system	2.0	11	2.4	13	5.6	30	0.676	0.03	**0.002**	0.19	**0.008**	0.16
Congenital malformations eye	1.1	6	2.0	11	1.5	8	0.217	0.08	0.583	0.03	0.487	0.03
Congenital malformations ear	1.9	10	1.3	7	1.5	8	0.463	0.04	0.635	0.03	0.795	0.02
Chromosome abnormalities	2.8	15	3.5	19	3.2	17	0.486	0.15	0.720	0.02	0.735	0.02
Diabetes	1.1	6	0.4	2	0.2	1	0.156	0.09	0.058	0.13	0.563	0.04
Epilepsy	3.7	20	4.3	23	5.4	29	0.641	0.03	0.189	0.08	0.394	0.05
Gastrointestinal and liver diseases	1.7	9	1.3	7	0.7	4	0.614	0.03	0.163	0.04	0.363	0.06
Heart - and blood diseases	2.2	12	2.6	14	1.5	8	0.691	0.02	0.367	0.05	0.196	0.08
Migraine or chronic headache	7.1	38	14.7	79	1.3	7	**0.000**	0.25	**0.000**	0.31	**0.000**	0.56
Muscular diseases	0.6	3	1.1	6	0.2	1	0.315	0.06	0.316	0.06	0.058	0.13
Musculoskeletal diseases												
Diseases of the back	2.2	12	2.8	15	2.2	12	0.559	0.04	1.000	0.00	0.559	0.04
Inflammatory polyarthropathies	0.6	3	0.7	4	0.4	2	0.705	0.02	0.654	0.03	0.413	0.05
Diseases of neck, shoulder and upper extremities	2.2	12	5.9	32	1.3	7	**0.002**	0.19	0.247	0.07	**0.000**	0.26
Psoriasis[c]	0.0	0	1.1	6	0.2	1	**0.014**	0.21	0.317	0.09	0.058	0.13
Mental chronic diseases												
Attention deficit/hyperactivity disorder (ADHD)	16.7	90	22.8	123	16.5	89	**0.012**	0.15	0.935	0.01	**0.009**	0.16
Autistic disorder	3.5	19	10.8	58	2.2	12	**0.000**	0.28	0.202	0.08	**0.000**	0.37
Dyslexia	5.9	32	14.1	76	1.5	8	**0.000**	0.28	**0.000**	0.25	**0.000**	0.53
Pervasive developmental disorder not otherwise specified (PDD-NOS)	11.5	62	13.9	75	10.2	55	0.235	0.07	0.493	0.04	0.061	0.11

[a] h = Cohen's h
[b] Bold differences are statistically significant

We also analyzed the dyad parents-GPs. GPs reported statistically significant higher prevalence rates for congenital malformations of the nervous system compared to the parents. However, the effect size was trivial. Parents reported statistically significant higher prevalence rates for six chronic diseases compared to the GPs. The effect sizes were trivial for chronic eczema and ADHD, small for diseases of neck, shoulder and upper extremities and autistic disorder, and moderate for migraine or chronic headache and dyslexia.

4.3.2. Concordance between the informants

Table 3 shows that in about half of all sixty six dyads concordance was poor to fair. In ten of all dyads the kappas ranged from good to very good. This occurred four times in the dyad teachers-parents, four times in the dyad parents-GPs and two times in the dyad teachers-GPs. In nine cases, it concerned somatic chronic diseases and in one case a mental chronic disease.

4.4. Discussion

This study shows that the concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents was relatively low. In about half of all 66 dyads the concordance was at most fair and just in 10 dyads good to very good.

4.4.1. Fit with previous studies

To our knowledge, no previous studies have examined the knowledge of teachers on the presence of a wide range of chronic diseases in ID-adolescents, and compared this with the knowledge parents and GPs have. However, we can state that our findings are mostly in line with studies reporting on a lack of information among teachers on issues associated with chronic diseases. These studies showed that teachers have limited knowledge regarding chronic disease-related information and limited confidence on their ability to work with children and adolescents with chronic diseases [18, 19, 23-25, 35]. Moreover, our results are in line with studies on inter-informant agreement. These studies also reported limited concordance between different informants. Disconcordance may be due to variations in situation of informants and due the nature of the problems: parents versus teachers, internalizing versus externalizing problems, visible versus not visible problems [36-38].

Table 3: Concordance between the knowledge of the three informants on the presence of chronic diseases in ID-adolescents (n=539) measured by Cohen's kappa.

	Teachers-Parents	Teachers-GPs	Parents-GPs
Somatic chronic diseases	Cohen's kappa	Cohen's kappa	Cohen's kappa
Asthma, chronic bronchitis, COPD	0.35	0.24	0.39
Cerebral palsy	0.57	0.33	0.28
Chronic eczema	0.27	0.26	0.29
Congenital malformations circulatory system	0.53	0.11	0.35
Congenital malformations nervous system	**0.74**[a]	0.42	0.45
Congenital malformations eye	0.22	0.01	0.09
Congenital malformations ear	0.58	0.55	0.53
Chromosome abnormalities	**0.82**	**0.87**	**0.83**
Diabetes	0.25	0.28	**0.67**
Epilepsy	0.59	0.55	**0.72**
Gastrointestinal and liver diseases	0.24	0.30	0.17
Heart - and blood diseases	0.53	0.39	0.44
Migraine or chronic headache	0.24	0.11	0.10
Muscular diseases	0.44	0.50	0.28
Musculoskeletal diseases			
Diseases of the back	**0.67**	0.49	0.51
Inflammatory polyarthropathies	0.57	**0.80**	**0.67**
Diseases of neck, shoulder and upper extremities	0.33	0.10	0.09
Psoriasis	-[b]	-	-
Mental chronic diseases			
Attention deficit/hyperactivity disorder (ADHD)	0.59	0.59	0.57
Autistic disorder	0.30	0.44	0.20
Dyslexia	0.39	0.03	0.00
Pervasive developmental disorder not otherwise specified (PDD-NOS)	**0.67**	0.51	0.53

[a] Bold kappas are good to very good

[b] Value of kappa was not meaningful to report because teachers did not report any case of psoriasis in ID-adolescents and GPs just one case of psoriasis in ID-adolescents

Our study shows that teachers reported mostly lower prevalence rates of chronic diseases in ID-adolescents than their parents, in particular on mental chronic diseases. In addition, this study shows that teachers and GPs and parents and GPs also reported different prevalence rates of chronic diseases in ID-adolescent. Anyhow, information on chronic diseases in ID-adolescents is available among different informants and the disagreement between them reflects different points of view between the informants and probably indicates a lack of communication between them.

4.4.2. Strengths and limitations

Important strengths of this study are that it examined the prevalence rates of a wide range of chronic diseases in ID-adolescents in a school-based sample representative for about 90% of the adolescents with ID. Another strength is the use of multi-informant information and the comparison of the similarly obtained data and the high response rates of teachers and GPs in our sub sample. A limitation is the relatively low response rate of the total sample (48%). This could have lead to selection bias, but non-response analyses revealed no major differences in response by age, gender and educational level.

4.4.3. Implications for practice

Teachers need to be fully aware of the presence of chronic diseases in ID-adolescents and the impact on their functioning in order to meet their needs and to support them successfully in the transition from school to work. They need professional advice to handle the problems of adolescents with ID and chronic diseases in their classroom [18, 19, 24, 25, 35]. Information from parents or healthcare professionals on the presence of chronic diseases in ID-adolescents could improve teachers' awareness of chronic diseases in ID-adolescents and the impact on their functioning, and could improve their confidence to support them adequately. However, arrangements must be found to improve the communication among teachers, parents and GPs to combine the knowledge and information on the presence of chronic diseases in ID-adolescents [18, 19, 24, 39, 40]. Parents are crucial in this communication process because they are in the position to inform both teacher and GP about the health condition of their child. Moreover, teachers and GPs need their informed consent to transfer information on the health condition of the adolescent.

4.4.4. Implications for research

Our study is the first to examine the knowledge of teachers, parents and GP's on the presence of a wide range of chronic diseases in ID-adolescents. Therefore, our findings need confirmation including an assessment of the validity of the knowledge of each informant. Moreover, future research should examine whether arrangements to improve the communication among teachers, parents and GPs will be effective [18]. Our results show very high prevalence rates of chronic diseases among ID-adolescents, and thus a high need for informed and skilled teachers to support these ID-adolescents in their school career and in their transition from school to work.

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Chapter 5

Emotional and behavioural problems in adolescents with intellectual disability with and without chronic diseases

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Abstract

Adolescents with intellectual disability (ID-adolescents) and adolescents with chronic diseases are both more likely to have emotional and behavioural problems. The aim of this study was to assess the association between chronic diseases in ID-adolescents and emotional and behavioural problems in a large school-based sample. We obtained data on 1044 ID-adolescents, aged 12-18, attending secondary schools in the Netherlands. Parents of the adolescents completed the Dutch version of the Strengths and Difficulties Questionnaire (SDQ) and questions about chronic diseases in their child and about the background of the child. Prevalence rates of emotional and behavioural problems were generally high in ID-adolescents with chronic diseases (45%), compared with ID-adolescents without chronic diseases (17%). The likelihood of emotional and behavioural problems were high in ID-adolescents with two (OR 4.47; 95% CI: 2.97-6.74) or more than two chronic diseases (OR 8.01; 95% CI: 5.18-12.39) and for ID-adolescents with mental chronic diseases (OR 4.56; 95% CI: 3.21-6.47). Also ID-adolescents with somatic chronic diseases had a high likelihood of emotional and behavioural problems (OR 1.99; 95% CI: 1.33-2.99), in particular in the combination of somatic and mental chronic diseases (OR 5.16; 95% CI: 3.46-7.71). The current study showed that chronic diseases in ID-adolescents, in particular mental chronic diseases, largely increase the likelihood of emotional and behavioural problems. This should be taken in the provision and planning of care for ID-adolescents.

5.1. Introduction

Adolescents with somatic or mental chronic diseases in the general population have a higher risk of emotional and behavioural problems compared with their healthy peers [1-11]. In addition, adolescents with intellectual disability (ID-adolescents) also have a higher risk of emotional and behavioural problems compared with their peers without ID [12-16]. Only a few studies examined the occurrence of emotional and behavioural problems in adolescents who had both problems, i.e. ID and one or more chronic diseases [17-26]. These studies are focused on either: (1) the presence or absence of a somatic chronic disease or physical complaints without taking into account the number and nature of the chronic diseases [20, 21]; or (2) one specific chronic diseases such as epilepsy, autism, cerebral palsy or ADHD without taking into account the presence of other chronic diseases [17-19, 22-26].

It is important to determine whether there is an association between the number and nature of chronic diseases and emotional and behavioural problems in ID-adolescents. An association, if found, would enable early intervention and treatment strategies by allowing identification of individuals who are at greater risk of emotional and behavioural problems. Preventing mental health problems in ID-adolescents and adequate treatment of those with mental health problems is important because mental health problems have a profound effect on the participation in educational programs, occupational opportunities, and the potential to live in the community, i.e. they are a major cause of failure of participation in the society [13, 27-29]. Finally, it is essential for the planning of interventions and services aimed at prevention or treatment.

The aim of this study is to assess: (1) the prevalence of emotional and behavioural problems in ID-adolescents with and without chronic disease; (2) the impact of chronic diseases in ID-adolescents on the increase of the likelihood of emotional and behavioural problems.

5.2. Methods

5.2.1. Participants and procedure

We collected data on adolescents with a borderline, mild, moderate or severe ID aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people). Nearly all adolescents of the target population attended secondary schools (schools for practical training) or special secondary schools (regional expertise centres). ID-adolescents

attending schools for practical training can be classified as mainly educable and have IQs between 60 and 80. ID-adolescents attending regional expertise centres can be classified as mainly trainable and have IQs between 30 and 59 [12]. ID-adolescents not attending secondary schools, most of them with profound ID, were not included

In the current school-based cross-sectional research project, 88% of the secondary schools (schools for practical training) or special secondary schools (regional expertise centres) in both provinces participated. Non-participating schools did not differ from participating schools regarding urbanization of the catchment area and number of students. All parents of the 2156 adolescents aged 12-18 years were sent a questionnaire and a reminder when they did not respond. One thousand forty four parents returned the questionnaire (48%). Adolescents in the response and non-response group did not differ regarding age (t-test=1.751, ns), but the response group had a higher proportion of girls ($\chi 2 = 5.9$; $p < 0.05$) and a higher proportion of adolescents with borderline or mild ID ($\chi 2 = 9.8$; $p < 0.05$). However, the effect sizes for both variables were negligible; Cohen's *w* were 0.06 and 0.07, respectively [30].

The study protocol was approved by the Medical Ethics Committee of the University Medical Centre Groningen, the Netherlands.

5.2.2. Measures

5.2.2.1. Intellectual disability

The target population had been officially classified as having ID by an independent committee established by the Dutch Ministry of Education, Culture and Science[31]. The classification of ID is based on a set of objective criteria, with the Dutch version of the Wechsler Intelligence Scale for Children-3rd Edition [32, 33], and the Snijders-Oomen Nonverbal intelligence test-Revised [34] as core ones.

5.2.2.2. Chronic diseases

Chronic diseases in ID-adolescents were measured by the National Permanent Survey on Living Conditions questionnaire (POLS); module health and labour, part chronic diseases in children [35]. POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic system; and ADHD. Questions were added about the presence of pervasive developmental disorders (PDD). Parents were asked to fill in the presence or absence of specific chronic diseases like for example: diabetes, asthma, epilepsy, and pervasive developmental disorder-not

otherwise specified, in the last twelve months in their children. Parents also had the possibility to mention the presence of chronic diseases that were not listed in the questionnaire.
POLS is developed by Statistics Netherlands and periodically used in a representative sample (n≈10.000) of the Dutch population [36].

5.2.2.3. Emotional and behavioural functioning

Emotional and behavioural functioning was assessed by the validated Dutch version of the Strengths and Difficulties Questionnaire (SDQ) [37-39]. The SDQ consists of 25 symptom items describing positive and negative attributes of children and adolescents. The 25 items are divided between five scales: emotional problem, conduct problem, inattention-hyperactivity, peer problem and prosocial behaviour. Each item has to be scored on a 3-point scale with 0='not true', 1='somewhat true', and 2='certainly true'. Sub-scale scores can be computed by summing scores on relevant items (after recoding reversed items; range 0–10). Higher scores on the prosocial behaviour sub-scale reflect strengths, whereas higher scores on the other four sub-scales reflect difficulties. A total difficulties score can also be calculated by summing the scores on the emotional symptoms, conduct problems, hyperactivity-inattention, and peer problems sub-scales (range 0–40) [40]. Several studies have shown the good reliability and validity of the SDQ in a non-ID population [41, 42]. Recently the SDQ is also used in studies with children and adolescents with ID to measure their emotional and behavioural functioning [14, 43-45].

5.2.2.4. Background characteristics

The questionnaire comprised the following questions on background characteristics: age, gender and school type of the adolescent. School type was used as proxy for severity of ID.

5.2.3. Analysis

The SDQ scores were dichotomised at the English 90th percentile cut-off of the sub-scales and total difficulties scale and the percentages of ID-adolescents with elevated scores was calculated [46]. Subsequently, adolescents were categorised in a group with and without chronic diseases. Next, we delimited the chronic diseases group regarding the number of chronic diseases: one, two and more than two (with a maximum of seven), and the nature of the chronic diseases: somatic (e.g. epilepsy, diabetes, asthma, etc.), mental (autism, pervasive developmental disorder not otherwise specified, dyslexia and attention deficit hyperactivity

disorder), or a combination of somatic and mental chronic diseases. Multivariate logistic regression analyses were performed and odds ratios (OR) were calculated, adjusted for level of ID (as measured by school type), to compare the groups pairwise and to identify groups of ID-adolescents with an increased likelihood for an elevated score on the SDQ scales

5.3. Results

Table 1 shows the background characteristics of the adolescents. The gender ratio, 58.2% boys and 41.8% girls, was similar to the ratio boys and girls with ID in the Netherlands [47].

Table 1: Demographic characteristics of the sample

Characteristics	
Age (n=1028)	Years
Mean	15.4
SD	1.6
Range	12-18
Gender (n=1035)	n (%)
Boys	602 (58.2)
Girls	433 (41.8)
Level of ID[a] (n=1038)	n (%)
IQ 60-80	785 (75.6)
IQ 30-59	253 (24.4)

[a]as measured by school type

5.3.1. With versus without chronic disease

Sixty one per cent (n=1044) of the ID-adolescents had at least one chronic disease. ID-adolescents with chronic diseases were more likely to have an elevated score on the SDQ problem behaviour scales and total difficulties scale than ID-adolescents without chronic diseases (Table 2).

Table 2: Percentages and odds ratio (OR), adjusted for level of ID, with 95% confidence interval (95% CI) of ID-adolescents with and without chronic diseases scoring above the cut-off on the SDQ scales.

	Chronic diseases		With versus without diseases[b]	
SDQ scales	Yes n=639	No n=397	OR	(95% CI)
Total difficulties	44.5%	17.3%	**2.54**[a]	**(1.87;3.44)**
Emotional problems	33.5%	16.4%	**1.69**	**(1.24;2.31)**
Conduct problems	25.9%	17.3%	**4.12**	**(2.89;5.87)**
Inattention-hyperactivity	31.9%	10.0%	**2.37**	**(1.80;3.13)**
Peer problems	46.2%	25.2%	**3.70**	**(2.75;4.97)**
Prosocial behaviour problems	14.3%	10.2%	1.45	(0.99;2.14)

[a]Bold results are statistically significant p <0.001.
[b]All level of ID effects p >0.05, except for Total difficulties scale p <0.005 and Peer problems scale p <0.000.

5.3.2. Number and nature of chronic diseases

ID-adolescents with one or more chronic diseases were more likely to have an elevated score on the problem behaviour scales and total difficulties scale than ID-adolescents without chronic diseases, conduct problems being the only exception (Table 3). ID-adolescents with more than two chronic diseases were most likely to score above the cut-off of the problem behaviour scales and total difficulties scale. They also had about a two fold higher likelihood to score above the cut-off on the prosocial behaviour scale, indicating problems in prosocial behaviour. With regard to the nature of chronic diseases, ID-adolescents with mental chronic diseases were as expected more likely to have an elevated score on the problem behaviour scales, prosocial behaviour scale and total difficulties scales compared with ID-adolescents without chronic diseases (Table 4). ID-adolescents with only somatic chronic diseases had a two fold higher likelihood to score above the cut-off on the emotional problem scale and total difficulties scale compared with ID-adolescents without chronic disease. ID-adolescents with a combination of somatic and mental chronic diseases had the highest likelihood to score above the cut-off on the problem behaviour scales and total difficulties scales compared with ID-adolescents without chronic diseases.

Table 3: Percentages and odds ratio (OR), adjusted for level of ID, with 95% confidence interval (95% CI) of adolescents without chronic diseases and with 1, 2, or > 2 chronic diseases scoring above the cut-off on the SDQ scales.

	Number of chronic diseases				0 versus 1[b]		0 versus 2[b]		0 versus 3[b]	
SDQ scales	0 n=397	1 n=305	2 n=164	>2 n=170	OR	95% CI	OR	95% CI	OR	95% CI
Total difficulties	17.3%	33.8%	49.3%	64.4%	**2.42**[a]	**(1.73;3.41)**	**4.47**	**(2.97;6.74)**	**8.01**	**(5.18;12.39)**
Emotional problems	16.4%	24.6%	38.9%	48.5%	**1.67**	**(1.16;2.39)**	**3.23**	**(2.13;4.92)**	**4.79**	**(3.12;7.36)**
Conduct problems	17.3%	20.1%	27.1%	38.3%	1.23	(0.85;1.78)	**1.81**	**(1.16;2.82)**	**3.05**	**(1.97;4.71)**
Inattention-hyperactivity	10.0%	24.9%	31.3%	48.9%	**2.99**	**(2.00;4.47)**	**4.01**	**(2.56;6.41)**	**8.27**	**(5.19;13.17)**
Peer problems	25.2%	36.0%	50.7%	64.7%	**1.64**	**(1.19;2.27)**	**2.84**	**(1.90;4.24)**	**4.58**	**(2.99;7.02)**
Prosocial behaviour problems	10.2%	13.6%	11.8%	18.8%	1.39	(0.89;2.16)	1.16	(0.64;2.10)	**1.98**	**(1.16;3.42)**

[a]Bold results are statistically significant p <0.001.
[b]All level of ID effects p > 0.05, except for Total difficulties scale p < 0.05 and Peer problems scale p < 0.000.

Table 4: Percentages and odds ratio (OR), adjusted for level of ID, with 95% confidence interval (95% CI) of adolescents without chronic diseases (0) and with somatic (S), mental (M) or somatic and mental (S&M) chronic diseases scoring above the cut-off on the SDQ scales.

	Nature of chronic diseases				0 versus S[b]		0 versus M[b]		0 versus S&M[b]	
SDQ scales	0 n=397	S n=244	M n=222	S&M n=173	OR	95% CI	OR	95% CI	OR	95% CI
Total difficulties	17.3%	30.0%	49.2%	53.8%	**1.99**[a]	**(1.33;2.99)**	**4.56**	**(3.21;6.47)**	**5.16**	**(3.46;7.71)**
Emotional problems	16.4%	26.7%	30.8%	45.6%	**1.85**	**(1.22;2.80)**	**2.26**	**(1.56;3.27)**	**4.21**	**(2.80;6.30)**
Conduct problems	17.3%	14.4%	29.6%	33.3%	0.82	(0.51;1.33)	**2.04**	**(1.41;2.94)**	**2.41**	**(1.59;3.66)**
Inattention-hyperactivity	10.0%	13.9%	39.3%	40.9%	1.42	(0.85;2.41)	**5.77**	**(3.86;8.61)**	**5.93**	**(3.80;9.26)**
Peer problems	25.2%	31.7%	49.6%	57.2%	1.29	(0.87;1.90)	**2.85**	**(2.03;3.99)**	**3.46**	**(2.33;5.12)**
Prosocial behaviour problems	10.2%	7.2%	19.8%	13.8%	0.68	(0.36;1.29)	**2.16**	**(1.39;3.35)**	1.37	(0.79;2.38)

[a]Bold results are statistically significant p <0.001
[b]All level of ID effects p >0.05, except for Total difficulties scale p <0.005 and Peer problems scale p <0.000

5.4. Discussion

Our results show that emotional and behavioural problems have a significantly higher prevalence in ID-adolescents with chronic diseases than in ID-adolescent without chronic diseases. The likelihood of emotional and behavioural problems increased by number of chronic diseases and was highest in ID-adolescents with two or more than two chronic diseases. Not surprisingly, ID-adolescents with mental chronic diseases had a high likelihood of emotional and behavioural problems. However, ID-adolescents with somatic chronic diseases also had a high likelihood of emotional and behavioural problems; in combination with mental chronic diseases they had the highest likelihood of emotional and behavioural problems.

5.4.1. Fit with previous studies

With respect to somatic chronic diseases, Dekker and Koot [21] studied a similar group of children and adolescents in the Netherlands. They found that children and adolescents with ID and somatic chronic diseases had a two and a half fold higher likelihood (OR 2.5) on Diagnostic and Statistical Manual of Mental Disorders-IV disorders compared with their peers without somatic chronic disease. This suggests that risks for parent-reported problems and for diagnosed disorders are increased among ID-adolescents with chronic diseases to a rather similar degree. However, Cormack et al. [20] found that children and adolescents with ID and severe physical disability had significantly lower mean scores on the Developmental Behaviour Checklist (DBC) primary carer version total score, the disruptive – and anxiety sub-scales. The properties of the instrument are likely to explain these differences. The DBC has been specifically designed to tap the emotional and behavioural problems of ID children and may therefore be expected to measure more precise the presence or absence of these problems among ID adolescents than a more general instrument such as the SDQ. Lewis et al. [23] used the DBC in a cohort study of children and adolescents with ID representative of the general Australian population of young people with ID. They found that children and adolescents with epilepsy did not differ from their non-epileptic peers in their level of behavioural or emotional disturbance. However, a potential source of error in their results was the accuracy of the parents to recall whether their children had epilepsy or not. Buelow et al. [19] used the Child Behaviour Checklist (CBCL) in a sample of children and adolescents with epilepsy. They found that 68% of the children and adolescents with ID and epilepsy and 51% of the children and adolescents without ID and epilepsy scored above the cut-off on the CBCL total

problem. Our results are in line with their results. Although items and scales of the SDQ and CBCL may have differential relevance among groups, comparable results are found in many populations [41]. Turky et al. [26] used the SDQ in a community-based population of children and adolescents with epilepsy. They found that children and adolescents with epilepsy and cognitive impairments had a higher likelihood to score above the cut-off on SDQ conduct problems (OR 14.78), inattention-hyperactivity (OR 9.44) and peer problems (OR 30.08) compared with children and adolescents with epilepsy and without special educational needs. Finally, Parkes et al. [24] used the SDQ in a sample based on population-based registers of cerebral palsy in eight European regions. They found that ID (IQ < 70) was associated with a significantly increased likelihood (OR 3.2) to score above the SDQ total difficulties cut-off in children with cerebral palsy. In both studies [24, 26] the higher odds ratios compared with the odds ratios in our study could be explained by the fact that they included adolescents without ID as reference group, whereas in the current study ID-adolescents without chronic diseases were the reference group.

With respect to mental chronic diseases, Bradley et al. [17] and Hill and Furniss [22] found that a higher proportions of children, adolescents and young adults with ID and autism scored above the cut-off on sub-scales of the diagnostic assessment for the severely handicapped-II (DASH-II) compared with their peers with ID and without autism. The results we found in the current study were in line with their results. However, it is of importance to mention that the DASH-II is in contrary to the SDQ specifically developed for individuals with severe or profound ID or developmental disabilities. Brereton et al. [18] used the DBC primary carer version in a sample drawn from the Australian Child and Adolescent Development Study, a longitudinal study of psychopathology in a representative sample of children and adolescents with intellectual disability in an Australian region. They found that children and adolescents with autism, 87% with ID, scored higher on the total behaviour problems mean score of the DBC primary carer version compared to children and adolescents with ID but without autism. Our results are in line with the results of Brereton et al. [18] but it should be noted that the DBC is specifically developed for children and adolescents with ID, whereas SDQ is not. Group-specific instruments can in general be expected to tap more of the problems that such a group has than instruments that are more general. Finally, Pearson et al. [25] used the Personality Inventory for Children-Revised (PIC-R) and found that children and adolescents with ID and ADHD had a higher risk to score above the cut-off on externalizing and internalizing problem behaviour of the PIC-R than their peers with ID but without ADHD. Our results confirm this mostly but a full comparison is difficult because Pearson et al. [25] reported about an undefined population of

children and adolescents with ID. In addition, the scope of PIC-R is broader than the scope of SDQ, as the PIC-R also assesses cognitive development.
Compared to other studies that reported prevalence rates of emotional and behavioural problems between 35% and 61% in children and adolescents with ID [12-16], we found a large difference in prevalence rates of emotional and behavioural problems in ID-adolescents. In the current study 17% of the ID-adolescents without chronic diseases and 30% to 64% of the ID-adolescents with chronic diseases had emotional and behavioural problems.

5.4.2. Strengths and limitations

This is the first study that examined the association between combinations of chronic diseases in adolescents with ID and emotional and behavioural problems in detail. A large school-based sample representative for about 95% of the adolescents with ID was used. A limitation of the study was the relatively low response rate (48%), but non-response analysis revealed that adolescents in the response and non-response group did not differ on age, gender and educational level. Next to this, adolescents with ID not attending school were not included, which may have influenced the prevalence of emotional and behavioural problems. Finally, the SDQ may underestimate the emotional and behavioural problems in ID-adolescents. First, because SDQ was not developed specifically for this group [48]. Second, non-verbal ID-adolescents may score as having fewer difficulties, because they cannot verbally lie or express headaches. Interpretation of this behaviour by the parent is more difficult compared with verbal ID-adolescents. However, because the population of non-verbal adolescents attending secondary schools in the Netherlands is very limited, it is unlikely that this will affect the results as we reported to an important degree.

5.4.3. Implications for clinicians

Our findings highlight the need for clinicians to be aware of the increased risk of emotional and behavioural problems in ID-adolescents who also have chronic diseases and that this risk potentially increases with the number of chronic diseases. Early identification and treatment of emotional and behavioural problems in this population is likely to also improve their well-being and social participation [13, 27-29]. The SDQ may provide a simple means of identifying emotional and behavioural problems in ID-adolescents since it is widely used in preventive child healthcare in the Netherlands [39].

5.4.4. Implications for research

Additional research is needed on the causal mechanisms behind the association of emotional and behavioural problems, and chronic diseases among ID-adolescents. This association may be due either to common causes for both, or to the chronic diseases leading to emotional and behavioural problems. These two mechanisms may lead to different strategies for early treatment. To disentangle these mechanisms, longitudinal research design studies are needed[13, 49]. Moreover, our findings regarding the association of ID severity and chronic diseases with emotional and behavioural problems conflict with previous ones that use the DBC. Additional research is needed to explain these differences.

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Chapter 6

Pervasive developmental disorder behaviour in adolescents with intellectual disability and co-occurring somatic chronic diseases

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Abstract

Evidence on the association between somatic chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour (PDD behaviour) is scarce. The aim of the present study is to assess the association between somatic chronic diseases in ID-adolescents and mild PDD behaviour. We obtained data on 1044 ID-adolescents, aged 12–18, attending secondary schools in the Netherlands. Parents of the adolescents completed the Dutch version of the Children's Social Behaviour Questionnaire (CSBQ) parent version, covering a wide range of PDD behaviour, and questions about chronic diseases and background characteristics of their child. ID-adolescents with somatic chronic diseases showed more PDD behaviour, in particular milder forms, than their peers without chronic diseases. In addition, ID-adolescents with somatic chronic diseases in combination with pervasive development disorders (PDD) and attention deficit hyperactivity disorder (ADHD) also showed more PDD behaviour than their peers with only PDD/ADHD. Clinicians should be extra alert on PDD behaviour, in particular the milder forms, in ID-adolescents when somatic chronic diseases are present. However, to strengthen our results about the relationship between somatic chronic diseases in ID-adolescents and PDD behaviour studies are needed using both the CSBQ and standardized diagnostic instruments.

6.1. Introduction

Studies on the association between somatic chronic diseases in adolescents with intellectual disability (ID-adolescents) and the full range of pervasive developmental disorder behaviour (PDD behaviour), in particular milder forms of PDD behaviour (mild PDD behaviour), are hardly available. Mild PDD behaviour is a term reserved for those who do not meet the criteria for severe PDD behaviour like in autism or Asperger syndrome. Mild PDD behaviour is widespread among ID-adolescents and has a profound effect on their daily functioning [1-4].

Literature shows a positive association between severity of ID and mild PDD behaviour in adolescents [1]. This positive association is caused by the fact that ID-adolescents, especially those with lower levels of ID, have a greater chance on both pervasive development disorders (PDD) [1, 5-7] and attention deficit hyperactivity disorder (ADHD) [6, 8]. Although ADHD and PDD have different nosological diagnoses, both diagnostic categories partly include similar symptoms like deficits in social interaction, impulsivity, attention and hyperactivity deficit [9]. These deficits are also associated with mild PDD behaviour [1, 3, 10].

Literature also suggests that adolescents with somatic chronic diseases show more PDD behaviour than adolescents in the general population [11-18], but only two studies have focused explicitly on ID-adolescents with somatic chronic diseases [16, 17]. None of the aforementioned studies used instruments that were suitable for screening or diagnosing mild PDD behaviour [3].

Studies on the association between somatic chronic diseases in ID-adolescents and PDD behaviour, in particular milder forms of PDD behaviour, are highly needed. Professionals do not always recognize mild PDD behaviour in (ID-) adolescents [2, 15]. Evidence on an association between somatic chronic diseases in ID-adolescents and mild PDD behaviour may thus increase the attentiveness of professionals for mild PDD behaviour, enabling earlier diagnosis and treatment. PDD behaviour is very disabling in social and interpersonal situations and hinder successful participation in society [1, 19, 20].

The aim of this study is to assess the association between somatic chronic diseases in ID-adolescents and PDD behaviour, in particular the milder forms of PDD behaviour.

6.2. Methods

6.2.1. Participants and procedure

We collected data in 2006-2007 from adolescents with a borderline, mild, moderate, or severe ID aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people). Nearly all adolescents of the target population attended secondary schools (schools for practical training) or special secondary schools (regional expertise centres). ID-adolescents attending schools for practical training can be classified as mainly educable and have IQs between 60 and 80. ID-adolescents attending regional expertise centres can be classified as mainly trainable and have IQs between 30 and 59 [21]. ID-adolescents not attending secondary schools, most of them with profound ID, were not included.

In the current school-based cross-sectional research project, 88% of the schools for practical training and regional expertise centres in both provinces participated. Non-participating schools did not differ from participating schools regarding urbanization of the catchment area and number of students. All parents of the 2156 adolescents aged 12-18 years received a questionnaire and a reminder when they did not respond. One thousand forty four parents returned the questionnaire (48%). Adolescents in the response and non-response group did not differ regarding age (t-test=1.751, ns), but the response group had a higher proportion of girls ($\chi 2 = 5.9$; $p < 0.05$) and a higher proportion of adolescents with borderline or mild ID ($\chi 2 = 9.8$; $p < 0.05$). However, the effect sizes for both variables were negligible; Cohen's *w* were 0.06 and 0.07, respectively [22].

The study protocol was approved by the Medical Ethics Committee of the University Medical Centre Groningen, the Netherlands.

6.2.2. Measures

6.2.2.1. Intellectual disability

The target population had been officially classified as having ID by an independent committee established by the Dutch Ministry of Education, Culture and Science [23]. The classification of ID is based on a set of objective criteria, with the Dutch version of the Wechsler Intelligence Scale for Children-3rd Edition [24, 25], and the Snijders-Oomen Nonverbal Intelligence Test-Revised [26] as core ones.

6.2.2.2. Chronic diseases

Chronic diseases in ID-adolescents were measured by the National Permanent Survey on Living Conditions questionnaire (POLS); module health and labour,

part chronic diseases in children [27]. POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic systems and ADHD. Questions were added about the presence of pervasive developmental disorders (PDD). Parents were asked to fill in the presence or absence of each specific chronic disease in the last 12 months for their child. Parents were also offered the possibility to mention the presence of chronic diseases that were not listed in the questionnaire. POLS was developed by Statistics Netherlands and is yearly used in a representative sample (n≈10.000) of the Dutch population [28].

6.2.2.3. Pervasive development disorder behaviour

PDD behaviour was measured by the Dutch version of the Children's Social Behaviour Questionnaire (CSBQ) parent version. The CSBQ has 49 items describing a broad range of behavioural features, including milder forms that are typical of PDD. The items can be allocated to six subscales: 'not optimally tuned to the social situation' (not tuned behaviour; 11 items addressing emotional overreacting and stubbornness/disobedience), 'reduced contact and social interest' (social withdrawn; 12 items), 'orientation problems in time, place, or activity' (orientation problems; 8 items), 'difficulties in understanding of social information' (not understanding; 7 items), 'stereotyped behaviour' (stereotyped behaviour; 8 items), and 'fear of and resistance to changes' (fear of changes; 3 items). Each item can be marked as 'does not apply' to the child (score 0), 'sometimes or somewhat applies' (score 1), or 'clearly or often applies' (score 2) [1, 3, 29]. Subscale scores can be computed by summing scores on the items; range 0–6 (fear of changes) to 0–24 (social withdrawn). Lower scores on the subscales reflect milder forms of PDD behaviour, whereas higher scores reflect more severe forms of PDD behaviour. A total score can also be calculated by summing the scores of the subscales (range 0–98). Although the CSBQ was originally developed for and investigated in children with normal intelligence, the psychometric qualities of the CSBQ in children with ID were found to be good [30, 31].

6.2.2.4. Background characteristics

The questionnaire comprised questions on age, gender and school type of the adolescent. School type was used as proxy for severity of ID.

6.2.3. Analysis

The CSBQ item scores were transformed to summative scores on the six subscales and a total CSBQ score, respectively. Subsequently, adolescents were categorised in a group without chronic diseases, with only somatic chronic diseases, with only PDD/ADHD, and with somatic chronic diseases in combination with PDD/ADHD. Adolescents with other psychiatric disorders (n=102) were excluded from the analyses.

We tested via Univariate Analysis of Variance with Bonferroni post-hoc correction the mean differences between the group with somatic chronic diseases versus without chronic diseases and the mean differences between the group with somatic chronic diseases in combination with PDD/ADHD versus the group with PDD/ADHD. Both analyses were adjusted for level of ID (as measured by school type). For all mean differences, effect sizes (Cohen's *d*) were calculated [22].

6.3. Results

Table 1: Demographic characteristics of the sample

Characteristics	
Age (n=1028)	Years
Mean	15.4
SD	1.6
Range	12-18
Gender (n=1035)	n (%)
Boys	602 (58.2)
Girls	433 (41.8)
Level of ID[a] (n=1038)	n (%)
IQ 60-80	785 (75.6)
IQ 30-59	253 (24.4)

[a]as measured by school type

Table 1 shows the background characteristics of the adolescents. The gender ratio, 58.2% boys and 41.8% girls, was similar to the ratio boys and girls with ID in the Netherlands [32].

Tables 2 and 3 shows the mean scores and standard deviations of the four subgroups of ID-adolescents on all CSBQ scales.

Table 2 shows that, adjusted for level of ID, ID-adolescents with somatic chronic diseases had statistically significant higher mean scores on all CSBQ scales compared to ID-adolescents without chronic diseases. According to Cohen's criteria, all the effect sizes were small, except for those associated with the Orientation problem scale; medium effect size.

Table 2: Means and standard deviations on CSBQ scales and mean differences, adjusted for level of ID, and effect sizes between CSBQ scale scores in ID-adolescents without chronic diseases and ID-adolescents with somatic chronic diseases.

Chronic diseases	None			SCD			MD[a]	p	95% CI	Cohen's d[c]
CSBQ scales	n	M	SD	n	M	SD				
Total score	392	15.27	12.89	233	21.95	14.66	**-6.53**	**<0.001**	**(-9.67;-3.40)**[b]	0.49
Not tuned behaviour	389	4.34	4.33	232	5.48	4.80	**-1.09**	**0.043**	**(-2.17;-0.02)**	0.25
Social withdrawn	392	2.76	3.56	233	4.29	4.23	**-1.51**	**<0.001**	**(-2.44;-0.57)**	0.40
Orientation problems	383	2.65	2.81	232	4.18	3.40	**-1.51**	**<0.001**	**(-2.21;-0.81)**	0.50
Not understanding	391	3.89	3.23	231	5.22	3.45	**-1.30**	**<0.001**	**(-2.02;-0.57)**	0.40
Stereotyped behaviour	389	0.88	1.71	231	1.59	2.19	**-0.68**	**0.006**	**(-1.22;-0.13)**	0.37
Fear of changes	388	0.68	1.12	229	1.17	1.59	**-0.48**	**0.001**	**(-0.82;-0.15)**	0.37

[a] All level of ID effects p<0.001
[b] Bold results are statistically significant
[c] Cohen thresholds: negligible effect (≤0.20); small effect (≥0.20 and <0.50); medium effect (≥0.50 and <0.80); large effect (≥0.80)
Note. *CI,* confidence interval; *MD,* mean differences; *SCD,* somatic chronic diseases

Table 3: Means and standard deviations on CSBQ scales and mean differences, adjusted for level of ID, and effect sizes between CSBQ scale scores in ID-adolescents with only PDD/ADHD and ID-adolescents with somatic chronic diseases in combination with PDD/ADHD.

Chronic diseases	PDD/AHDH			SCD+PDD/ADHD			MD[a]	p	95% CI	Cohen's d[c]
CSBQ scales	n	M	SD	n	M	SD				
Total score	163	33.82	16.60	144	41.03	16.62	**-7.21**	**<0.001**	**(-11.55;-2.88)**[b]	0.44
Not tuned behaviour	163	9.92	5.71	144	10.38	5.46	-0.46	1.000	(-1.95;1.02)	0.08
Social withdrawn	163	6.50	5.16	144	8.59	5.35	**-2.08**	**<0.001**	**(-3.38;-0.79)**	0.40
Orientation problems	163	5.73	3.47	144	7.32	3.72	**-1.59**	**<0.001**	**(-2.55;-0.63)**	0.44
Not understanding	161	7.34	3.70	144	8.44	3.35	**-1.10**	**0.024**	**(-2.10;-0.09)**	0.31
Stereotyped behaviour	162	2.45	3.06	143	3.73	3.83	**-1.28**	**<0.001**	**(-2.02;-0.53)**	0.37
Fear of changes	163	1.90	1.87	143	2.49	1.97	**-0.59**	**0.005**	**(-1.05;-0.13)**	0.31

[a] All level of ID effects p<0.001
[b] Bold results are statistically significant
[c] Cohen thresholds: negligible effect (≤0.20); small effect (≥0.20 and <0.50); medium effect (≥0.50 and <0.80); large effect (≥0.80)
Note. *CI,* confidence interval; *MD,* mean differences; *SCD,* somatic chronic diseases

In addition, Table 3 shows that, adjusted for level of ID, ID-adolescents with comorbidity of somatic chronic diseases in combination with PDD/ADHD had statistically significant higher mean scores on five of the six CSBQ subscales and total CSBQ score compared to ID-adolescents with only PDD/ADHD. The effect sizes were small for all of the significant associations.
As shown in both Tables 2 and 3, all level of ID effects were statistically significant. This indicates that ID-adolescents with IQ between 30 and 59 had higher mean scores on CSBQ subscales and total CSBQ score compared to ID-adolescents with IQ between 60 and 80.

6.4. Discussion

ID-adolescents with somatic chronic diseases have more PDD behaviour than those without chronic diseases, in particular milder forms of PDD behaviour. This association is independent of having PDD/ADHD or not. Our findings suggest a relationship between somatic chronic diseases in ID-adolescents and mild PDD behaviour.
Differences are rather large, whereas effect sizes are mostly relative low. This can be explained by the fact that the differences in mean scores on the CSBQ scales between the groups as well as the deviations of the means within each group are considerably large, indicating considerably variation within each group.

6.4.1. Fit with previous studies

To our knowledge, no previous studies have examined the association between somatic chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour (PDD behaviour), including milder forms of PDD behaviour. However, our findings are mostly in line with two previous studies on ID-adolescents that used instruments suitable for diagnosing severe forms of PDD behaviour and reported prevalence rates of PDD based on diagnostic classification [16, 17]. Both studies used the Autism Behaviour Checklist (ABC) and Childhood Autism Rating Scale (CARS) and found higher prevalence rates of autistic spectrum disorder in ID-adolescent with epilepsy (38%) [17], cerebral palsy (11%) [16] and severe visual impairment (50%) [16]. Moreover, our findings are also mostly in line with studies on autism spectrum disorders and somatic chronic diseases among adolescents without ID and among a mixed group of adolescents with and without ID [11, 13-15, 18]. These studies showed that adolescents with somatic chronic diseases had a greater chance on autism spectrum disorders compared to adolescents in the general population [12].

Our study thus shows that findings on severe cases of PDD can be extended to the full range of PDD behaviours.

6.4.2. Strength and limitations

The strength of this study is that it examined the association between somatic chronic diseases in adolescents with ID and PDD behaviour in a large school-based sample representative for about 95% of the adolescents with ID. Another strength of this study was the use of a specifically developed screening instrument that covers the full range of PDD behaviour, including milder forms. The CSBQ is useful for describing the severity and pattern of social deficits in groups other than PDD. A limitation is that the CBSQ is not intended for purposes of diagnostic classification [3]. Another limitation of the study was the relatively low response rate (48%), but non-response analyses revealed that adolescents in the response and non-response group did not differ on age, gender and educational level.

6.4.3. Implications for clinicians

Clinicians should be extra alert on PDD behaviour, in particular the milder forms, in ID-adolescents when somatic chronic diseases are present. Detecting PDD behaviour in ID-adolescents and adequate treatment of those with mild PDD behaviour is important because these problems are very disabling for ID-adolescents in social and interpersonal situations and hinder successful participation in the society [1, 19, 20]. The CSBQ may be helpful as a first screening device when there is a suspicion of PDD behaviour, even when the problems are associated with diagnoses outside autism spectrum disorders. In these cases, the score profile of the adolescent may help to plan for more specific diagnostic assessment and treatment, as well as their monitoring [3].

6.4.4. Implications for research

Our study was the first to assess the association between somatic chronic diseases in ID-adolescents and mild PDD behaviour. Therefore, they need confirmational studies, which preferable use both the CSBQ and standardized diagnostic instruments.

6.5. Acknowledgement

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Chapter 7

Main findings, discussion and implications

7.1. Introduction

The aims of this thesis were: (1) to explore the prevalence of chronic diseases in adolescents with intellectual disability (ID-adolescents) and the impact on their behaviour; and (2) to assess the knowledge about chronic diseases in ID-adolescents among teachers working in schools for practical training and special secondary schools. These goals were translated into the following five main research questions:

1. What is known in the literature on the prevalence rates of chronic diseases in populations of children and adolescents with ID?
2. What is the prevalence of chronic diseases in ID-adolescents aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people)?
3. What is the concordance between the knowledge teachers have on the presence of chronic diseases in ID-adolescents and the knowledge of parents and healthcare professionals?
4. What is the association between chronic diseases in ID-adolescents and their emotional and behavioural functioning?
5. What is the association between chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour?

7.2. Main findings

7.2.1 Research question 1: What is known in the literature on the prevalence rates of chronic diseases in populations of children and adolescents with ID?

A systematic literature review was conducted to explore the prevalence rates of chronic diseases in ID-adolescents. The systematic literature review showed high prevalence rates of a wide range of chronic diseases in children with ID. The six most prevalent chronic diseases in children with ID were epilepsy (22.0%), cerebral palsy (19.8%), any anxiety disorder (17.1%) oppositional defiant disorder (12.4%), Down syndrome (11.0%) and autistic disorder (10.1%). The prevalence rates of chronic diseases in children with ID were disconcertingly higher than the prevalence rates of chronic diseases reported in studies on children without ID.

7.2.2. Research question 2: What is the prevalence of chronic diseases in ID-adolescents aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people)?

A study on the prevalence rates and the nature of chronic diseases in ID-adolescents was conducted. This study showed that about 63% of the ID-adolescents had at least one chronic disease. The six most prevalent chronic diseases were: ADHD (21.1%), PDD-NOS (14.0%), dyslexia (13.9%), migraine or chronic headache (12.7%), autistic disorder (10.9%) and asthma, chronic bronchitis, COPD (9.9%). Gender was not associated with the number of chronic diseases, but boys were more likely than girls to be diagnosed with mental chronic diseases and with a combination of somatic and mental chronic diseases. Severity of ID was positively associated with the number of chronic diseases. With regard to the type of the chronic diseases, adolescents with moderate and severe ID were more likely than adolescents with mild ID to be diagnosed with chronic disease regarding any somatic chronic diseases, any mental chronic diseases and regarding a combination of somatic and mental chronic diseases. In addition, adolescents with severe ID were more likely than adolescents with moderate ID to be diagnosed with a combination of somatic and mental chronic diseases.

The prevalence rates of chronic diseases in ID-adolescents that were found in this study were compared with prevalence rates of chronic diseases among adolescents in the general population. We found that for 8 of the 17 chronic diseases assessed, the prevalence rates were statistically significant higher among ID-adolescents than among adolescents in the general population. Differences were particularly large with regard to mental chronic diseases (ADHD, autistic disorder, and PDD-NOS) and small for somatic diseases.

7.2.3. Research question 3: What is the concordance between the knowledge teachers have on the presence of chronic diseases in ID-adolescents and the knowledge of parents and healthcare professionals?

In this study the knowledge of teachers on the presence of chronic diseases in ID-adolescents was assessed and compared with the knowledge parents and GPs have on the presence of chronic diseases in these adolescents. Teachers reported predominantly lower prevalence rates of chronic diseases in ID-adolescents than parents, in particular on mental chronic diseases. In addition, this study showed that teachers and GPs and parents and GPs also reported different prevalence rates of chronic diseases in ID-adolescents. Moreover, the concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents was relatively low. In about half of all 66 dyads the

concordance was for the most part fair and just in 10 dyads good to very good; nine of these latter cases concerned somatic chronic diseases.

7.2.4. Research question 4: What is the association between chronic diseases in ID-adolescents and their emotional and behavioural functioning?

This study focused on the association between chronic diseases in ID-adolescents and their emotional and behavioural functioning. Prevalence rates of emotional and behavioural problems were generally high in ID-adolescents with chronic diseases, compared with ID-adolescents without chronic diseases. The likelihood of emotional and behavioural problems was high in ID-adolescents with two or more than two chronic diseases and for ID-adolescents with mental chronic diseases. In addition, ID-adolescents with somatic chronic diseases had also a high likelihood of emotional and behavioural problems, in particular regarding the combination of somatic and mental chronic diseases. Both analyses were adjusted for level of ID (as measured by school type).

7.2.5. Research question 5: What is the association between chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour?

This study focused on the association between chronic diseases in ID-adolescents, in particular the somatic ones, and the full range of PDD behaviour. Its results showed that ID-adolescents with somatic chronic diseases had more PDD behaviour - in particular milder forms of PDD behaviour - than adolescents without chronic diseases. This association is independent of having PDD/ADHD or not. The analysis was adjusted for level of ID (as measured by school type).

7.3. Methodological considerations

This paragraph discusses the strengths and limitations of this thesis and the procedures used in the systematic literature review and the four empirical studies.

7.3.1. Systematic literature review

The systematic literature review is the first inclusive study on the prevalence of chronic diseases in children with ID. The main strength of this systematic literature review is its comprehensive search strategy including all relevant literature databases on a broad range of chronic diseases, including a check of the references from all the studies retrieved.

Some limitations in the systematic literature review should also be considered. First, with regard to the search strategy it is possible that due to inadequate indexing some studies included in the databases were not retrieved. In order to minimise this retrieval bias, a manual search of references was a key part of the search process. Second, though the reviewers valued the quality of the included studies from good to high, the prevalence rates for most chronic diseases varied widely between the included studies. Several factors that were not examined and appraised in this study can contribute to this variation. For example, the accessibility of healthcare for children with ID in different countries can influence the characteristics of the sample; also the quality of registers can influence the prevalence rates of chronic diseases that were found in the included studies. Moreover, the method of diagnosis, diagnoses retrieved from clinical examination versus extracted from registers, and the use of different classification frameworks, DSM versus ICD, can influence the results of the included studies, also explaining some of the differences in prevalence rates [1-5].

7.3.2. Empirical studies

An important strength of the empirical studies was the use of large samples, community -and school-based, and representative for ID-adolescents. Other strengths were the examination of the prevalence rates of a wide range of chronic diseases in ID-adolescents and the comparison of most outcomes with similarly collected data on adolescents without ID.

A limitation of the empirical studies was the relatively low response rate (48%) among parents, but non-response analyses revealed that the response and non-response group did not differ on adolescents' age, gender and educational level. Another limitation may have been the use of SDQ and CSBQ questionnaires because originally these are developed for and investigated in children with normal intelligence. However, in line with the results of previous research in children with ID [6-12], the internal consistency of the SDQ total score and hyperactivity scale and the CSBQ total score and subscales in this study were good (Cronbach's alphas > 0.70). The SDQ emotional problem -, conduct problem -, peer problem - and prosocial behaviour subscales had Cronbach's alphas between 0.61 and 0.69. Finally, a limitation may have been the difficulty experienced by parents in filling out the questionnaire, in particular by those with lower levels of cognitive performances. However, functionally illiterate parents may have been non-responders and in cases where parents were legally incapable, their legal representative received the questionnaire.

Finally, due to the cross-sectional design of the empirical studies causal statements on the relation between the different variables could not be made.

7.4. Discussion of main findings

This thesis originates from the need of schools for practical training and schools for special secondary education to know more about the chronic diseases of their pupils with ID, and the impact of these diseases on emotional and behavioural functioning of these pupils - including the full range of PDD behaviour.
Schools for practical training and schools for special secondary education prepare pupils to participate in society and to enter the labour market [13]. It is important to determine: whether and to what degree ID-adolescents have additional chronic diseases, teachers' knowledge on the presence of these diseases, and the impact of chronic diseases on the adolescents' emotional and behavioural functioning. Both ID and chronic diseases can lead to limitations in their capacities. Capacities seem to be particularly limited in case of emotional and behavioural problems. In addition, limited capacities can have a profound effect on participation of ID-adolescents in educational programs and occupational opportunities and societal participation [14-24]. However, evidence on the prevalence rates of chronic diseases in ID-adolescents, teachers' knowledge on the presence of these diseases, and the impact of chronic diseases on the adolescents' emotional and behavioural functioning was limited till now.

7.4.1. Prevalence of chronic diseases

This thesis shows that the prevalence rates of chronic diseases in ID-adolescents is disconcertingly high compared to adolescents without ID, in particular with regard to mental chronic diseases. Four factors have been suggested in literature to explain these high rates. First, biological/genetic factors, i.e. genetic and chromosomal disorders could cause both ID and a wide range of chronic somatic and mental diseases. Second, chronic diseases, in particular those which affect brain functioning can lead to ID. Third, the association may simply reflect the point of a general underlying relationship between cognitive performance and problem behaviour associated with mental chronic diseases. For example problems in social functioning are apparent in autism spectrum disorders and ID [25, 26]. Fourth, the association of ID with social, environmental and economic factors such as poverty, poor housing and neighbourhood conditions or living in a residential centre because of ID, risky behaviour, prenatal exposure to toxins (alcohol and drugs), under-nutrition, may lead to adverse health outcomes and to higher rates of chronic diseases [18, 26, 27].

7.4.2. Emotional and behavioural functioning, including the full range of PDD behaviour

This thesis also shows a positive association between emotional and behavioural problems in ID-adolescents and chronic diseases; in particular mental chronic diseases. However, somatic chronic diseases, in particular in combination with mental chronic diseases, are also associated with higher levels of emotional and behavioural problems.

The association between chronic diseases and emotional and behavioural problems in ID-adolescents may be explained by similar mechanisms as the association between ID and chronic diseases. First, emotional and behavioural problems are likely to be caused by biological/genetic factors, in particular those who affect brain functioning. Second, the association may simply reflect the point of a general underlying relationship between emotional and behavioural problems and diagnostic characteristics associated with certain mental chronic diseases. Third, adolescents' and family factors are also associated with emotional and behavioural problems. Adolescents' dysfunction, e.g. due to developmental disorders or reduced coping skills are likely to cause emotional and behavioural problems. Health problems of the parent(s) and reduced coping skills of the parent(s) are also likely to cause emotional and behavioural problems in ID-adolescents. Fourth, emotional and behavioural problems may also partially be attributed to an increased risk of exposure to adverse socio-economic circumstances such as low education level of the parents, reduced household income or living in poor neighbourhoods [17, 27-33].

Moreover, our findings suggest a relationship between somatic chronic diseases in ID-adolescents and the full range of PDD behaviour, in particular the milder forms. This relationship has not been assessed in previous studies [34, 35]. A reason for this may be that instruments suitable for screening or diagnosing mild PDD behaviour were only just developed recently [36].

7.4.3. Teachers' knowledge on chronic diseases

Finally, this thesis shows that the knowledge of teachers working in schools for practical training and in special secondary schools on the health status of their pupils can be improved. Even though several informants have information on chronic diseases in ID-adolescents, this information is not always shared. The disagreement between parents, teachers and healthcare professionals may be due to variations in situation of informants and reflects probably a lack of communication among them on chronic diseases in their child, pupil and patient, respectively [37-39]. This confirms earlier findings that teachers' knowledge about

chronic disease-related information of their pupils is far below what is necessary. It may be due to the fact that parents and healthcare professionals usually do not share this kind of information with teachers [14, 16, 21, 40].

7.5. Implications for professional practices and policymakers

This thesis enlarges the knowledge on the presence of chronic diseases in ID-adolescents, and on factors that are associated with these conditions: high levels of emotional and behavioural problems. In addition, this thesis shows that the concordance between the knowledge of teachers, parents and GPs on the prevalence of chronic diseases in ID-adolescents is relatively low. These findings should alert professionals and policymakers to these problems.

7.5.1. Teachers

Schools for practical training and schools for special secondary education play a pivotal role in the challenging process to teach their pupils, i.e. ID-adolescents, competences to enter the labour market and to participate successfully in society. The findings as presented may have implication for teachers at these schools.

The increased risk on chronic diseases and on emotional and behavioural problems - including the full range of PDD behaviour - in ID-adolescents should alert teachers working in schools for practical training and schools for special secondary education. Preventing these health problems in ID-adolescents and adequate treatment of ID-adolescents with these problems are important because of their profound effects on the participation of ID-adolescents in educational programs and subsequently their occupational opportunities [14-21, 23, 24, 40-47].

Teachers need information on health problems because it can improve their ability to work with ID-adolescents. The knowledge of teachers may be improved by enhancing the communication between them and parents and healthcare professionals. Parents are crucial in this communication process because they are in the position to inform both teacher and healthcare professionals about the health condition of their child; professionals need their informed consent to transfer information on the adolescent's health condition. Arrangements must be developed to improve the communication among teachers, parents and healthcare professionals [14, 16, 21, 40].

Information on chronic diseases and emotional and behavioural problems are core elements in the total assessment of the pupil. Literature indicates that the assessment of ID-adolescents should integrate elements on health, vocational

interest and academic -, social - and vocational capacities. Schools should add relevant questions to their intake form to explore the presence of chronic diseases and emotional and behavioural problems and enter these topics on the agenda in regularly talks with parents and pupils. They can also use validated questionnaires such as the SDQ or CSBQ to obtain information from parents on the occurrence of these problems in their child. In addition, they can approach parents for informed consent in order to retrieve relevant information from healthcare professionals.
The information obtained by teachers and a variety of health and social services professionals on the ID-adolescents' limitations and capacities can be the input for the development of a detailed care and transition plan, comprising health, academic, social and vocational needs. This plan will support teachers and ID-adolescents in the classroom and subsequently the ID-adolescents' transition to work and societal participation. The success of this trajectory also depends on: the competences of teachers to work with children and adolescents with disabilities, continuing education of teachers on these topics, arrangements such as case- or care management to coordinate the efforts from different professionals, and the use of evidenced-based interventions in daily practice [15, 21, 48-52].

7.5.2. Healthcare professionals

Healthcare professionals need data on the prevalence on chronic diseases and emotional and behavioural problems - including the full range of PDD behaviour - in ID-adolescents for the early detection and adequate treatment of these conditions and to prevent the burden of these conditions in ID-adolescents and their families [22, 23, 53-59]. In the Netherlands GPs and preventive child healthcare (PCH) professionals are crucial in the identification and registration of chronic diseases and associated problems. Dutch residents consult their GP first if they have a health problem. Consequently, doctor-defined health problems as presented to GPs in the Netherlands should provide a valid profile of morbidity in the population [60, 61]. However, this thesis shows that the concordance between the knowledge of parents and GPs on the prevalence of chronic diseases in ID-adolescents is relatively low. This may indicate that GPs have insufficient knowledge on the presence of chronic diseases in ID-adolescents. Their knowledge could be improved by enhancing the communication with parents and ID-adolescents and by their putting the topic on the agenda for regular talks with them and/or preventive home visits. They could also use screening instruments to detect health problems in ID-adolescents. Moreover, the cooperation and the transfer of information between GPs and other healthcare professionals, such as PCH professionals, could be improved [49, 62].

PCH provides health and developmental monitoring to all Dutch children aged 0-19. All children undergo three to four assessments by PCH professionals during their school careers, mostly in primary school. These assessments consist of checking a wide range of factors (developmental, physical and psychosocial) as part of the routine health monitoring. At the end of the assessment, the preventive child health physician decides whether there is any need for counselling, follow-up, or referral to other professionals or services. In general PCH professionals do not offer curative care but they do offer parenting support [6, 63]. A recent development in the Netherlands is the integration of PCH in Centres for Youth and Family. These centres will offer parents, children and young people aged up to 23 a recognizable, centralized, family-friendly helpdesk providing advice on parenting and all aspects of growing up, adequate and appropriate assistance, and coordination of the help given. The centres will also act as central notification points for professionals, warning them early about families in difficulty. Finally, these centres must encourage the cooperation and coordination between the different institutions (healthcare, welfare, social security services, police and judiciary). In 2011 Centres for Youth and Families should be operational in every municipality in the Netherlands [64-66].

During preparation of the research project PCH-organizations were asked to provide prevalence rates of chronic diseases in adolescents, however, no valid prevalence rates could be provided. One of the main reasons for this omission is that individual PCH professionals do not always share the same criteria when rating children's health; leading to large differences between PCH professionals with respect to the detection and the proportion of children identified as being (un)healthy [6, 63, 66, 67].

The results of this thesis lend support to the importance of policies that seek to screen for health problems among ID-adolescents. For example, previous research has shown that the use of validated questionnaires, training of PCH professionals in a structured method for identifying psychosocial problems, and providing more time and more frequent assessments in the adolescence phase may improve the accuracy of early identification of these problems [68, 69]. Although the SDQ nowadays is widely used in PCH in the Netherlands, the CSBQ may also be helpful as a first screening device when there is a suspicion of PDD behaviour, even when the problems are associated with diagnoses outside autism spectrum disorders. In these cases, the score profile of the adolescent may help to plan for more specific diagnostic assessment and treatment, as well as their monitoring [36]. Moreover, the cooperation and the transfer of information between PCH professionals and other healthcare professionals, such as GPs, should be improved [49, 62].

7.5.3. Policymakers

The Human Rights Conventions of the United Nations, in particular the Convention on the Rights of the Child (CRC) and the Convention on the Rights of Persons with Disabilities (CRPD) state that persons with disabilities, among which ID-adolescents, have the same right to participate in society as all other persons without any discrimination [70, 71].

Governments should recognize that ID-adolescents have special needs and the right to special care. They should take all appropriate measures to provide these services, and these services should be designed to ensure that ID-adolescents have effective access to and receive education, training, (preventive) health care services, rehabilitation services, preparation for employment and recreation opportunities in a manner conducive to the adolescents' achieving the fullest possible social integration and individual development [70, 71]. It should be noted that ratification of the Human Rights Conventions of the United Nations by governments gives them the obligation to support practices to take all appropriate measures to realize these rights. However, according to the concluding observations report of the Committee on the Rights of the Child in 2009 the rights of children in the Netherlands can be improved. The Committee recommends that the Dutch Government [72]:

1. Take all necessary measures to ensure that legislation providing protection for persons with disabilities, as well as programmes and services for children with disabilities, are effectively implemented;
2. Develop and strengthen early identification programmes and early intervention programmes;
3. Undertake awareness-raising campaigns on the rights and special needs of children with disabilities, encourage their inclusion in society and prevent discrimination and institutionalization;
4. Provide training for professional staff working with children with disabilities, such as medical, paramedical and related personnel, teachers and social workers;
5. Ratify the International Convention on the Rights of Persons with Disabilities (CRPD) and its Optional Protocol, signed on 30 March 2007.

The intention to ratify the CRPD by the Dutch government will give a positive incentive to policies realizing these rights [73]. This gives school and health practices the opportunity to make a claim to the government to provide them with sufficient resources. According to the former chairperson of the United Nations Committee

on the Rights of the Child (prof. dr. mr. J.E. Doek) practices can improve their actions towards the Dutch Government to remind them of their obligations to support them with all appropriate measures to realise these rights [74].

This thesis shows high prevalence rates of chronic diseases in ID-adolescents and associated high risks on emotional and behavioural problems. Moreover, this thesis shows that the knowledge of teachers and healthcare professionals on health problems in ID-adolescents could be improved. Literature indicates that the number of ID-adolescents is increasing in the Netherlands [52]. ID-adolescents have special needs that require health and related services of a type or amount beyond those required by adolescents generally. These health and related services (e.g. education, health - and social care) are needed to handle the huge burden of these conditions, both with regard to prevention and treatment in order to enhance the well-being and societal participation of ID-adolescents [22, 75]. However, there is evidence that professional care and support delivered to ID-adolescents can be improved because it is ineffective, fragmented, poorly coordinated and a large number of ID-adolescents and their parents experience unmet needs [49, 52, 76-78]. Moreover, occupational opportunities for ID-adolescents with and without chronic diseases in the Netherlands are not promising, even though employment-related benefits are available [52, 79, 80].

The growing numbers of ID-adolescents, their high risk of health problems in combination with their unmet needs should alert policymakers. The results of this thesis and what is known in literature on the care ID-adolescents need should be translated into policy measures. Policy makers should stimulate and finance effective care arrangements focused on the adolescents' development and social integration to tackle these problems. Literature indicates that these arrangements should be comprehensive and transdisciplinary. Key elements in these arrangements are effective early detection methods of health problems in ID-adolescents and effective treatment of these problems to alleviate the burden of these problems in ID-adolescents and their families [49, 52, 62]. Literature also indicates that continuous education, ideally multidisciplinary, on these topics improves the competences of professionals to work with children and adolescents with disabilities and the use of effective methods [49, 52, 62, 81, 82].

Policymakers should pay attention to incentives that facilitate the continuous improvement of competences of professionals. Moreover, policymakers should pay attention to incentives that are needed to promote effective communication and cooperation between professionals and the coordination of the effort of the different professionals, as the adolescents navigate across settings [15, 21, 48-52]. Case or care management have been suggested in literature as an innovative strategy

to realize this, but a discussion should be started on the responsibilities, roles and tasks of the different professionals and the financing of time spent on cooperation and case - or care management [49, 52, 83].

Much can be gained by improving policies that support professionals to enhance their daily practices. This will improve the success of ID-adolescents in educational programs and subsequently their occupational opportunities and societal participation.

7.5.4. Implications for research

This thesis reports on different studies and based on the findings and the methodological considerations recommendations can be made for future research.

First, evidence is needed on the prevalence studies of a wide range of chronic diseases in ID-adolescents. These studies should employ multiple data sources and should ideally use the same classification framework of diseases to improve the validity and comparability of the prevalence rates.

Second, the results of the study on the comparison of the knowledge of teachers, parents and GPs on the presence of chronic diseases in ID-adolescents need confirmation by other studies including an assessment of the validity of the knowledge of each informant. Future research also should examine whether arrangements to improve the communication among teachers, parents and GPs will be effective.

Third, the results of the study on the association between combinations of chronic diseases in adolescents with ID and emotional and behavioural problems need additional research to explore the causal mechanisms behind this association. This association may be due either to common causes for both, or to the chronic diseases leading to these problems. These two mechanisms may lead to different strategies for early treatment.

Fourth, the results of the study on the association between chronic diseases in adolescents with ID and the full range of PDD behaviour needs also confirmation by other studies, which preferably use both the CSBQ and standardized diagnostic instruments. Confirmation of this association would enable early intervention and treatment strategies by allowing identification of individuals who are at greater risk of PDD behaviour.

Finally, literature indicates that evidence on the effectiveness of the interventions in school and PCH practices is limited [16, 21, 52, 62]. Closer attention should be paid to the effectiveness of interventions focused on prevention, early identification of problems, interventions to tackle problems, or case management. Interventions that appear to be promising in helping parents to raise children, such as

Stepping Stones Triple P, should therefore be extensively evaluated in the Netherlands [52, 65, 84].

The recently developed Dutch Academic Collaborative Centres, like Care4Youth in the northern part of the Netherlands, will stimulate research on the effectiveness of these practices and the interventions professionals use. This will lead to the best possible care and support of ID-adolescents in their trajectory to participation in society.

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Summary

Chapter 1

Chapter 1 introduces the key concepts and the broader context of this thesis and presents its aims, research questions and the methodology of the studies.

The studies originate from the need of schools for practical training and schools for special secondary education to know more about the chronic diseases of their pupils with ID and about the impact of these diseases on their emotional and behavioural functioning.

Schools for practical training and schools for special secondary education prepare pupils for entering the labour market and for participation in society. They need information on whether and to what degree ID-adolescents have additional chronic diseases because both ID and chronic diseases can lead to limitations in adolescents' capacities. Capacities seem to be particularly limited in case of emotional and behavioural problems. In addition, limited capacities can have a profound effect on participation of ID-adolescents in educational programs, occupational opportunities and societal participation. However, evidence on the prevalence rates of chronic diseases in ID-adolescents and the impact on their emotional and behavioural functioning was limited.

The aims of this thesis were: (1) to explore the prevalence of chronic diseases in ID-adolescents and the impact on their behaviour; and (2) to assess the knowledge about chronic diseases in ID-adolescents among teachers working in schools for practical training and special secondary schools. These goals were translated in the following five research questions:

1. What is known in the literature on the prevalence rates of chronic diseases in populations of children with ID?
2. What is the prevalence of chronic diseases in ID-adolescents aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people)?
3. What is the concordance between the knowledge teachers have on the presence of chronic diseases in ID-adolescents and the knowledge of parents and healthcare professionals?
4. What is the association between chronic diseases in ID-adolescents and their emotional and behavioural functioning?
5. What is the association between chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behaviour?

Chapter 2

The second chapter provides a systematic literature review on the prevalence rates of chronic diseases in populations of children with ID.

The past decades have shown an increase in the knowledge on chronic diseases in children with ID. In several studies ID in children was associated with a range of chronic diseases. A major difficulty in studies on this subject is the wide variability of prevalence rates that are reported for specific chronic diseases in children with ID. As a result, the validity of prevalence rates has to be disputed. However, policymakers and professionals value and need valid prevalence rates. Policymakers need these data for the planning and financing of adequate care arrangements. Professionals need these data for the early detection and adequate treatment of chronic diseases and to alleviate the burden of these conditions in ID-adolescents and their families. The aim of this study was to provide a systematic literature review of the prevalence rates of chronic diseases in populations of children with ID in general.

We identified 2994 relevant studies by searching Medline, Cinahl and PsycINFO databases in the period 1996-2008. Finally, 31 studies with sufficient methodological quality were included. The reported prevalence rates of chronic diseases in children with ID were disconcertingly high and were much higher than in the general population. However, both the number of studies that were included and the number of chronic diseases they reported about were limited.

The systematic literature review showed that much can be gained in the caring for children with ID by better evidence on the occurrence of chronic diseases. Various recommendations for clinicians and researchers are discussed to improve this evidence.

Chapter 3

In this chapter the prevalence rates of a wide range of chronic diseases in ID-adolescents aged 12 – 18 years in two provinces in the north of the Netherlands, Groningen and Drenthe (total population of about 1.1 million people) were assessed. In addition, these prevalence rates were compared with the prevalence rates among adolescents in the general population.

Valid community-based data on the prevalence rates of the full range of chronic diseases in ID-adolescents were scarce. Only a few studies have reported such prevalence rates of a wide range of chronic diseases associated with ID. Moreover, only some studies compared their results with the prevalence rates of chronic diseases in the general population, but none of these studies reported prevalence rates of chronic diseases among ID-adolescents. The aim of this study was to

assess the prevalence rates and the nature of chronic diseases in a population of ID-adolescents and to compare them with rates among adolescents in the general population.
We obtained data on 1083 parents of ID-adolescents attending schools for practical training, special secondary schools, day care centres or living in residential centres, fully covering one region of the Netherlands. Disconcertingly high prevalence rates of a wide range of chronic diseases in ID-adolescents were found. Of all ID-adolescents about 63% had at least one chronic disease. For 8 of the 17 chronic diseases that we assessed the prevalence rates were statistically significantly higher among ID-adolescents than among adolescents in the general population. Differences in prevalence rates were larger if ID was more severe, regarding any somatic chronic diseases, any mental chronic diseases and regarding a combination of somatic and mental chronic diseases.
The results of this study show a need for effective care arrangements to handle this huge burden of morbidity, both regarding its prevention, and its treatment. As such, it provides a challenge to both clinicians and policy.

Chapter 4

In Chapter 4 the knowledge of teachers on the presence of chronic diseases in ID-adolescents was compared with the knowledge parents and healthcare professionals had on this subject.
Teachers need to be fully aware of the presence of chronic diseases in ID-adolescents and the impact on their functioning in order to meet their needs and to support ID-adolescents successfully in the transition from school to work. However, evidence on teachers' knowledge about somatic and mental chronic diseases among ID-adolescent compared to the knowledge parents and healthcare professionals have, was limited. The aim of this study was to assess the knowledge of teachers on the presence of chronic diseases in ID-adolescents and to compare teachers with parents and healthcare professionals and parents with healthcare professionals regarding their knowledge on the presence of chronic diseases in these ID-adolescents.
We obtained data on 1044 parents of ID-adolescents attending schools for practical training and special secondary schools, fully covering one region of the Netherlands. Teachers, parents and general practitioners (GPs) of the adolescents completed a questionnaire about the occurrence of chronic diseases.
Although prevalence rates of chronic diseases among ID-adolescents were very high, concordance between teachers, parents and health care professionals on the presence of chronic diseases in ID-adolescents was relatively low. In addition,

teachers reported mostly lower prevalence rates of chronic diseases in ID-adolescents compared to the parents. This confirms earlier findings that teachers' knowledge about chronic disease-related information of their pupils is far below what is necessary. The knowledge of teachers may be improved by enhancing the communication between them and parents and healthcare professionals. Arrangements must be developed to improve the communication among teachers, parents and healthcare professionals.

Chapter 5

Chapter 5 focused on the impact of chronic diseases in ID-adolescents on the increase of the likelihood of emotional and behavioural problems. Emotional and behavioural problems have a profound effect on participation in educational programs, occupational opportunities and the potential to participate in the community. ID-adolescents and adolescents with chronic diseases are both more likely to have emotional and behavioural problems. However, evidence on the occurrence of emotional and behavioural problems in adolescents who have both problems, i.e. ID and one or more chronic diseases, was scarce. The aim of this study was to assess the association between chronic diseases in ID-adolescents and emotional and behavioural problems.

We obtained data on 1044 ID-adolescents attending schools for practical training and special secondary schools, fully covering one region of the Netherlands. Parents of the adolescents completed the Dutch version of the Strengths and Difficulties Questionnaire (SDQ).

The current study showed that the number of chronic diseases in ID-adolescents largely increase the likelihood of emotional and behavioural problems. Not surprisingly, ID-adolescents with mental chronic diseases had a high likelihood of emotional and behavioural problems. However, ID-adolescents with somatic chronic diseases also had a high likelihood of emotional and behavioural problems; in combination with mental chronic diseases they had the highest likelihood of emotional and behavioural problems.

These findings highlight the need for clinicians to be aware of the increased risk of emotional and behavioural problems in ID-adolescents who also have chronic diseases and that this risk potentially increases with the number of chronic diseases. Early identification and treatment of emotional and behavioural problems in this population is likely to improve well-being and social participation.

Chapter 6

Chapter 6 focused on the impact of chronic diseases in ID-adolescents on PDD behaviour, in particular the association between somatic chronic diseases and milder forms of PDD behaviour.

PDD behaviour is very disabling for ID-adolescents in social and interpersonal situations and hinder successful participation in the society. Evidence on the association between somatic chronic diseases in ID-adolescents and the full range of PDD behaviour was scarce. The aim of this study was to assess the association between somatic chronic diseases in ID-adolescents and PDD behaviour, in particular the milder forms of PDD behaviour.

We obtained data on 1044 ID-adolescents attending schools for practical training and special secondary schools in the Netherlands. Parents of the adolescents completed the Dutch version of the Children's Social Behaviour Questionnaire (CSBQ) parent version.

The current study showed that ID-adolescents with somatic chronic diseases have more PDD behaviour than their peers without chronic diseases, in particular milder forms of PDD behaviour. This association is independent of having PDD/ADHD, or not.

These findings suggest a relationship between somatic chronic diseases in ID-adolescents and mild PDD behaviour. Clinicians should be extra alert on PDD behaviour, in particular the milder forms, in ID-adolescents when somatic chronic diseases are present. Early identification and treatment of PDD behaviour in this population is likely to improve their well-being and social participation. The CSBQ may be helpful as a first screening device when there is a suspicion of PDD behaviour, even when the problems are associated with diagnoses outside autism spectrum disorders.

Chapter 7

In chapter 7 each of the answers on the research question will be considered in relation to the aims of the thesis and the methodological considerations of the studies.

The aims of this thesis were: (1) to explore the prevalence of chronic diseases in ID-adolescents and the impact on their behaviour; and (2) to assess the knowledge about chronic diseases in ID-adolescents among teachers working in schools for practical training and special secondary schools.

This thesis shows high prevalence rates of chronic diseases in ID-adolescents and associated high risks on emotional and behavioural problems including the full range of pervasive developmental disorder behaviour (PDD behaviour). Moreover,

this thesis shows that the knowledge of teachers and healthcare professionals on health problems in ID-adolescents could be improved.
Based on the results of this thesis and what is known in literature recommendations for practice, policy and research are discussed in this chapter. The main recommendations focus on the following topics:

- The rights of the child
- Comprehensive and transdisciplinary care arrangements
- Early detection and effective treatment of health problems in ID-adolescents
- Communication between parents and professionals
- Cooperation between professionals
- Coordination of the efforts of the various professionals
- Continuous improvement of professional competences

Samenvatting

Hoofdstuk 1

In hoofdstuk 1 wordt de bredere context van het proefschrift en de begrippen die in het proefschrift centraal staan toegelicht.

Het proefschrift is ontstaan uit de behoefte van scholen voor praktijkonderwijs en scholen voor speciaal voortgezet onderwijs, de regionale expertisecentra's (RECs), om meer kennis te krijgen over het vóórkomen (prevalentie) van chronische aandoeningen bij hun leerlingen met een intellectuele beperking en de impact hiervan op emotionele en gedragsproblemen. Vanuit wetenschappelijke hoek was hier tot nu toe relatief weinig aandacht voor.

Scholen voor praktijkonderwijs en de RECs, in dit proefschrift betreft het de RECs 1-3, bereiden hun leerlingen voor op arbeidsmarkt en zelfstandig functioneren in de samenleving. Het hebben van een intellectuele beperking en één of meerdere chronische aandoeningen kunnen leiden tot beperking in de mogelijkheden van adolescenten om deel te nemen aan arbeidsmarkt en samenleving. De mogelijkheden worden nog eens extra beperkt indien adolescenten daarnaast ook nog emotionele en gedragsproblemen hebben.

Het proefschrift heeft de volgende doelstellingen:

1. Het vaststellen van de prevalentie van chronische aandoeningen bij adolescenten met een intellectuele beperking en de impact hiervan op emotionele en gedragsproblemen; en
2. Het vaststellen van de kennis van docenten die werkzaam zijn op scholen voor praktijkonderwijs en de RECs over de aanwezigheid van chronische aandoeningen bij hun leerlingen.

De doelstellingen zijn vertaald in de volgende vijf onderzoeksvragen:

1. Wat is in de literatuur bekend over de prevalentie van chronische aandoeningen bij kinderen met een intellectuele beperking?
2. Wat is de prevalentie van chronische aandoeningen bij adolescenten met een intellectuele beperking in de leeftijdscategorie 12-18 jaar in de provincies Groningen en Drenthe?
3. Welke overeenstemming is er tussen de kennis die docenten hebben over de aanwezigheid van een chronische aandoening bij hun leerlingen vergeleken met de kennis die ouders en de huisarts hierover hebben?
4. Welke samenhang is er tussen het hebben van een chronische aandoening bij adolescenten met een intellectuele beperking en emotionele en gedragsproblemen?

5. Welke samenhang is er tussen het hebben van een chronische aandoening bij adolescenten met een intellectuele beperking en gedrag passend bij pervasieve ontwikkelingsstoornissen (PDD gedrag), in het bijzonder de samenhang tussen somatische chronische aandoeningen en de lichtere vormen van PDD gedrag?

Hoofdstuk 2

In hoofdstuk 2 worden de resultaten beschreven van een systematische literatuur review naar de prevalenties van chronische aandoeningen bij kinderen met een intellectuele beperking.

De afgelopen decennia heeft een stijging laten zien van de kennis over de aanwezigheid van chronische aandoeningen bij kinderen met een intellectuele beperking. Verschillende onderzoeken tonen aan dat kinderen met een intellectuele beperking te maken kunnen krijgen met een brede range van chronische aandoeningen. Echter, de gevonden prevalenties van specifieke chronische aandoeningen verschillen aanzienlijk. Onduidelijk is in hoeverre de gevonden prevalenties valide zijn. Valide cijfers over de prevalentie van chronische aandoeningen bij kinderen met een intellectuele beperking zijn van groot belang voor zowel het beleid als voor zorgpraktijken. Beleidmakers hebben deze informatie nodig voor de planning en financiering van goede zorgarrangementen voor kinderen met een intellectuele beperking en hun families. Professionals in de zorg hebben deze informatie nodig voor het vroegtijdig opsporen en adequaat behandelen van chronische aandoeningen en het ondersteunen van kinderen met een intellectuele beperking, waardoor de negatieve gevolgen die zij kunnen ervaren als gevolg van de aandoeningen verminderd worden. Het doel van dit onderzoek was een systematische literatuur review inzake de prevalentie van chronische aandoeningen bij kinderen met een intellectuele beperking.

De volgende databases werden geraadpleegd naar artikelen betreffende de prevalentie van chronische aandoeningen bij kinderen met een intellectuele beperking in de periode 1996-2008: Medline, Cinahl and PsycINFO. De zoektocht leverde 2994 relevante artikelen op. Uiteindelijke voldeden 31 artikelen aan de vooraf vastgestelde methodologische criteria. De gevonden prevalenties van chronische aandoeningen bij kinderen met een intellectuele beperking in de 31 artikelen waren relatief zeer hoog. Echter, zowel het aantal artikelen als de chronische aandoeningen waarover gerapporteerd werd waren beperkt.

De systematische literatuur review toont aan dat er relatief vaak chronische aandoeningen voorkomen bij adolescenten met een intellectuele beperking vergeleken met adolescenten in de algemene populatie. Echter, er valt nog

veel te verbeteren aan de validiteit van deze gegevens. In dit hoofdstuk worden verschillende aanbevelingen gedaan om de validiteit van prevalentie cijfers van chronische aandoeningen bij kinderen met een intellectuele beperking te verbeteren.

Hoofdstuk 3

In hoofdstuk 3 wordt verslag gedaan van een onderzoek naar de prevalentie van chronische aandoeningen bij adolescenten met een intellectuele beperking (12-18 jaar) in de provincies Groningen en Drenthe.

De gevonden prevalentie cijfers werden vergeleken met cijfers over de prevalentie van chronische aandoeningen bij adolescenten in de algemene populatie.

Er zijn weinig valide op bevolkingsonderzoek gebaseerde cijfers over de prevalentie van een brede range van chronische aandoeningen bij mensen met een intellectuele beperking. Slechts een paar studies hebben prevalentie cijfers over een brede range van chronische aandoeningen gerapporteerd. Daarnaast worden deze prevalentie cijfers zelden vergeleken met prevalentie cijfers van chronische aandoeningen bij mensen in de algemene populatie. Bij adolescenten is een vergelijking nog nooit gemaakt. De prevalentie van chronische aandoeningen bij adolescenten met een intellectuele beperking in Groningen en Drenthe werd vastgesteld aan de hand van een vragenlijst die door de ouders werd ingevuld. De adolescenten werden geworven via scholen voor praktijkonderwijs, de RECs, centra's voor dagbesteding en instellingen voor mensen met een intellectuele beperking. In totaal is van 1083 ouders (respons 48%) de vragenlijst retour ontvangen. Bij 63% van hun kinderen bleken één of meerdere chronische aandoeningen aanwezig te zijn. Verder bleek dat 8 van de 17 onderzochte chronische aandoeningen significanter vaker voorkwamen bij adolescenten met een intellectuele beperking dan bij met adolescenten in de algemene populatie. De ernst van de intellectuele beperking bleek positief samen te hangen met het aantal chronische aandoeningen. Bovendien bleek dat met de ernst van de intellectuele beperking de kans toeneemt op het hebben van een somatische, een mentale en de combinatie van somatische en mentale aandoeningen.

De resultaten die in dit hoofdstuk beschreven worden geven aan dat adolescenten met een intellectuele beperking relatief vaak bijkomende chronische aandoeningen hebben, wat gepaard gaat met een relatief hoge behoefte aan adequate zorgarrangementen. Daar ligt een grote uitdaging voor professionals en beleidmakers.

Hoofdstuk 4

In hoofdstuk 4 wordt de kennis van docenten, werkzaam op scholen voor praktijkonderwijs en de RECs, over de aanwezigheid van chronisch aandoeningen bij hun leerlingen, i.c. adolescenten met een intellectuele beperking, vergeleken met de kennis die ouders en huisartsen van deze adolescenten hierover hebben.
Het is van groot belang dat leraren op de hoogte zijn van de aanwezigheid van chronische aandoeningen bij hun leerlingen om hen zo optimaal mogelijk te begeleiden naar de arbeidsmarkt. Echter, kennis hierover ontbreekt in de literatuur.
In totaal is van 1044 ouders van adolescenten met een intellectuele beperking die praktijkonderwijs of onderwijs aan de RECs in Groningen of Drenhte volgen een vragenlijst retour ontvangen (respons 48%). Zowel ouders, docenten als huisartsen vulden een lijst in betreffende de aanwezigheid van chronische aandoeningen bij de adolescent. Docenten, ouders en huisartsen rapporteerden relatief hoge prevalenties van chronische aandoeningen bij de adolescenten. Echter, de overeenstemming tussen hen over de aanwezigheid van chronische aandoeningen bij de adolescenten was relatief laag. Bovendien rapporteerden docenten in de meeste gevallen lagere prevalentie cijfers betreffende de aanwezigheid van chronische aandoening bij hun leerlingen dan de ouders van de leerlingen.
De bevindingen die gedaan zijn tijdens dit onderzoek bevestigen dat de kennis van docenten over de aanwezigheid van chronische aandoeningen en daaraan gerelateerde problemen verbeterd kan worden. Om dat te realiseren moeten arrangementen gevonden worden om de communicatie tussen ouders, docenten en huisartsen te verbeteren.

Hoofdstuk 5

Hoofdstuk 5 richt zich op de invloed die chronische aandoeningen bij adolescenten met een intellectuele beperking hebben op de aanwezigheid van emotionele en gedragsproblemen.
Emotionele en gedragsproblemen hebben een negatieve invloed op de participatie aan schoolprogramma's, op het krijgen en behouden van werk en uiteindelijk op het meedoen in de samenleving. Zowel adolescenten met een intellectuele beperking als die met een chronische aandoening hebben een grotere kans op emotionele en gedragsproblemen vergeleken met adolescenten die dit niet hebben. Echter, onderzoek naar het optreden van emotionele en gedragsproblemen bij adolescenten die zowel een intellectuele beperking als een chronische aandoening hebben is beperkt.
In totaal is van 1044 ouders van adolescenten met een intellectuele beperking die praktijkonderwijs of onderwijs aan de RECs volgen de Nederlandse versie

van de SDQ vragenlijst retour ontvangen (respons 48%). Het aantal chronische aandoeningen dat adolescenten hebben hangt positief samen met de kans emotionele en gedragsproblemen. Niet verrassend bleek dat vooral voor adolescenten met mentale chronische aandoeningen te gelden. Echter, ook indien adolescenten somatische chronische aandoeningen hadden bleek de kans op emotionele en gedragsproblemen verhoogd. Adolescenten met een combinatie van somatische en mentale chronische aandoeningen bleken de grootste kans te hebben op emotionele en gedragsproblemen. Deze bevindingen benadrukken dat professionals alert moeten zijn indien adolescenten met een intellectuele beperking bijkomende chronische aandoeningen hebben. Vroegtijdige onderkenning van emotionele en gedragsproblemen en een adequate behandeling vergroten de kans op een beter welbevinden en sociale participatie.

Hoofdstuk 6

Hoofdstuk 6 richt zich op de invloed die chronische aandoeningen bij adolescenten met een intellectuele beperking hebben op PDD gedrag, in het bijzonder op de samenhang tussen somatische chronische aandoeningen en de lichtere vormen van PDD gedrag.

PDD gedrag beperkt adolescenten met een intellectuele beperking in situaties waarin sociale vaardigheden van belang zijn, met als resultaat dat zij niet volledig kunnen participeren in de samenleving. Het wetenschappelijk bewijs aangaande de samenhang tussen somatische chronische aandoeningen en PDD is zeer gering. In totaal is van 1044 ouders van adolescenten met een intellectuele beperking die praktijkonderwijs of onderwijs aan de RECs volgen de Vragenlijst voor Inventarisatie van Sociaal gedrag van Kinderen (VISK, in het Engels CSBQ) vragenlijst retour ontvangen (respons 48%). Adolescenten met een intellectuele beperking en een somatisch chronische aandoening vertoonden meer PDD gedrag, in het bijzonder de lichtere vormen, dan adolescenten met een intellectuele beperking zonder chronische aandoeningen. Deze samenhang is ook aanwezig bij de groep adolescenten met een PDD/ADHD diagnose.

De bevindingen wijzen op een relatie tussen somatisch chronische aandoeningen en PDD gedrag. Professionals dienen extra alert te zijn op PDD gedrag bij adolescenten met een intellectuele beperking indien zij ook een somatisch chronische aandoening hebben. Vroegtijdige onderkenning van PDD gedrag en een adequate behandeling vergoten de kans op een beter welbevinden en sociale participatie. De VISK kan ondersteunend zijn voor professionals als screeningsinstrument voor het opsporen van PDD gedrag, zelfs indien het gaat om diagnoses die buiten het autisme spectrum vallen.

De resultaten beschreven in dit proefschrift tonen aan dat de prevalentie van chronische aandoeningen bij adolescenten met een intellectuele beperking zeer hoog is. De aanwezigheid van chronische aandoeningen gaat samen met een verhoogde kans op emotionele en gedragsproblemen, inclusief PDD gedrag. Bovendien tonen de resultaten beschreven in dit proefschrift aan dat de kennis die docenten en huisartsen hebben over de aanwezigheid van chronische aandoeningen bij adolescenten met een intellectuele beperking verbeterd kan worden.

In dit hoofdstuk worden verschillende aanbevelingen gedaan voor de praktijk, het beleid en wetenschappelijk onderzoek. De aanbevelingen zijn gebaseerd op de bevindingen in dit proefschrift en op wat bekend is in de literatuur. De belangrijkste aanbevelingen richten zich op:

- De rechten van kinderen
- Vroegtijdige onderkenning en behandeling van gezondheidsproblemen bij adolescenten met een intellectuele beperking
- Communicatie tussen ouder en professionals
- Communicatie tussen professionals
- Breed georiënteerde transdisciplinaire zorgprogramma's
- Coördinatie van de interventies van de verschillende professionals
- Een verbetering van de competenties van professionals

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CV

Barth Oeseburg werd geboren op 29 september 1961 te Groningen. Hij behaalde zowel zijn MAVO als HAVO diploma in Groningen, waarna hij in 1986 aan de HBO-V in Groningen is afgestudeerd. Hij werkte van 1987 tot en met 1993 als verpleegkundige op verschillende verpleegafdelingen van het Universitair Medisch Centrum Groningen (UMCG). Gelijktijdig startte hij de studie Sociologie aan de Rijksuniversiteit Groningen (RUG) en rondde deze in 1993 af. In 1994 werd hij de eerste verpleegkundig consulent Multiple Sclerose (MS) in Nederland werkzaam voor zowel de afdeling neurologie van het UMCG als het Coördinatiecentrum Chronisch Zieken Noord-Nederland (CCZ-NN), een project van de Nationale Commissie Chronisch Zieken. Hij startte verschillende zorgvernieuwingsprojecten, onder andere op het gebied van (poli)klinische zorg, transmurale zorg, thuiszorg en wonen en zorg voor jonge mensen met MS. Hij was verder mede-ontwikkelaar van de Farmaceutische patiëntenzorg-standaard MS van het Wetenschappelijk Instituut Nederlandse Apothekers. In 1997 werd hij projectleider van verschillende zorgvernieuwingsprojecten binnen het CCZ-NN, onder andere op het gebied transmurale zorg voor mensen met fibromyalgie en een coping training voor mensen met een chronische ziekte en hun partner in de thuiszorg. Voor verschillende projecten verwierf hij externe financiering via onder meer zorgverzekeraars en ZonMW. In de periode 2000-2006 was hij lid van de commissie van het praktijk en beleidsprogramma chronisch zieken van ZonMW.
In 2001 maakte hij de overstap naar wetenschappelijk onderzoek, RUG disciplinegroep gezondheidswetenschappen, waar hij eerst een kwalitatief onderzoek deed naar de verantwoordelijkheden en de verdeling daarvan in de zorg voor chronisch zieken, in het bijzonder mensen met MS (sectie metamedica, financiering door NWO programma Ethiek & Beleid). In 2005 was hij als onderzoeker/projectmedewerker verbonden aan het project preventie en prevalentie gefinancierd door ESF, programma EQUAL II (disciplinegroep Public Health Research / Toegepast Gezondheidsonderzoek). De data verzameld tijdens dit project hebben geleid tot dit proefschrift.
Vanaf 2008 is hij als opleidingscoördinator verpleegkundige vervolgopleiding chronisch zieken verbonden aan het UMCG Wenckebach Instituut, School of Nursing and Health.

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